SIBLINGS OF YOUTH WITH A DISABILITY

SIBLINGS OF YOUTH WITH A DISABILITY DURING THE TRANSITION TO ADULTHOOD: A SYNTHESIS OF RESOURCES AND UNDERSTANDING OF THEIR EXPERIENCES TO SUPPORT THEM IN CURRENT AND FUTURE ROLES

By

LINDA NGUYEN, B.H.Sc., M.Sc

A Thesis
Submitted to the School of Graduate Studies
In Partial Fulfillment of the Requirements for the Degree
Doctor of Philosophy in Rehabilitation Science

DOCTOR OF PHILOSOPHY (2022) McMaster University
(School of Rehabilitation Science) Hamilton, Ontario

TITLE:	Siblings of youth with a disability during the transition to adulthood: Understanding their experiences and synthesizing resources to support them in current and future roles
AUTHOR:	Linda Nguyen B.H.Sc (Honours) (McMaster University) M.Sc. (McMaster University)
SUPERVISOR:	Dr. Jan Willem Gorter, M.D., Ph.D.
PAGES:	xxii, 437

LAY ABSTRACT

As youth with a disability transition to adulthood, many will assume greater responsibility for their own health management, activities often supported by family members, including siblings. Little information is available about the roles that siblings have to their sibling with a disability in this process. Siblings have mentioned that they need information and resources for their roles. The purpose of this work is to summarize resources and programs in Canada and internationally for siblings, as well as better understand the experiences of siblings of an individual with a disability. The information from this work can be helpful by sharing the summary of resources with siblings and families, create new resources to support siblings of an individual with a disability, and build on programs that allow for siblings and the whole family to participate.

ABSTRACT

As youth with disabilities age and transition to adulthood, they will often need to learn how to manage their healthcare. Siblings are members of the family who can provide support to their sibling with a disability, but their roles during transition have not been well described in the literature. Typically developing (TD) siblings identified that they require information and resources in their roles.

The overall objectives of the research studies in this doctoral dissertation are: 1) to synthesize existing resources and programs to support TD siblings of individuals with a disability; and 2) to develop a deeper understanding about the experiences of TD siblings. The findings from these studies are summarized in six scholarly manuscripts prepared for peer review and publication. All studies were conducted in partnership with siblings with lived experiences.

Results from the review and qualitative document analysis of online resources to support TD siblings (Chapter 2) identified that limited resources are available to support TD siblings with healthcare management of their sibling with a disability. The scoping review (with the protocol described in Chapter 3 and results presented in Chapter 4) identified that programs for TD siblings are focused on knowledge development and skill acquisition for the TD siblings themselves or on empowerment by training TD siblings in skills that they can apply with their sibling with a disability. In addition to synthesizing existing knowledge, the qualitative study provided an in-depth understanding of the experiences of TD siblings (with the protocol described in Chapter 5) in two distinct areas: their roles and responsibilities (Chapter 6) and the influence of their sibling and family relationships on their identity formation during developmental stages (Chapter 7).

Overall findings from this dissertation suggest areas for future directions, including knowledge transfer to share synthesized resources with target audiences, co-creation of resources to support siblings in their roles, and enhancement of programs to involve siblings and the whole family.

ACKNOWLEDGEMENTS

It is surreal for me to think that I am at the "end" of my PhD journey after having worked on multiple projects in my patient-oriented doctoral research program in the School of Rehabilitation Science and CanChild Centre for Childhood Disability Research over the past few years. The completion of this dissertation would not have been possible without the incredible support, collaborations, and teamwork from multiple individuals in my life.

First, I am immensely grateful to my supervisor, Dr. Jan Willem Gorter. He has been so kind, caring, and supportive throughout this dissertation. He always encouraged me to have work-life balance, yet still challenged me to critically think about the next steps in my doctoral research program and career. I am appreciative of the guidance offered from my supervisory committee members: Dr. Susan Jack, Dr. Briano Di Rezze, and Dr. Marjolijn Ketelaar. Dr. Susan Jack has been an incredible mentor who motivates me to think critically about research methodologies and supported me in my personal life. Dr. Briano Di Rezze has always provided thoughtful considerations about the application of research to future work with suggestions of how to engage with community organizations. Dr. Marjolijn Ketelaar has been so kind and giving with her time, and I enjoyed our conversations about our reflections about partnering with young people and families in research along with ideas of future projects.

Thank you to my partnership with the Sibling Youth Advisory Council (SibYAC) including: Dana Arafeh, Samantha Bellefeuille, Hanae Davis, Julia Hanes, Jessica Havens, and James Koutrakos. I am honoured that you were so willing to share your stories and lived experiences as a sibling of an individual with a disability. All of you continuously inspire me to continue with research to increase awareness and include siblings in child health research. I am deeply thankful of our partnership.

Words cannot express the gratitude that I feel about the family that I have formed at CanChild and the School of Rehabilitation Science. Thank you to Tram Nguyen, Patrick McPhee, and Andrea Cross who provided advice about their doctoral and postdoctoral experiences. Thank you to my colleagues who were always available for a conversation: Sandra Abdel Malek, Jael Bootsma, Monika Novak Pavlik, Samantha Micsinzski, and Kinga Pozniak. I have gained incredible experiences under the guidance of the McMaster team in the READYorNot™ Brain-Based Disabilities Project: Barb Galuppi, Nadilein Mahlberg, and Sonya Strohm. I am also thankful to the staff at CanChild who were always willing to offer support when I had questions about my projects. Thank you, Dayle McCauley, for your guidance with grant applications; and thank you, Marlice Simon, for your support. I continue to learn and be inspired from my undergraduate students, Claire Dawe-McCord and Samantha Dong. I also appreciate that the CanChild family introduced me to other researchers and students in the field internationally. In particular, thank you to Bente van Oort for your inspiring and encouraging me to reflect on partnerships with young people. Thank you to my mentors, Dr. Julie Richardson, Dr. Peter Rosenbaum, and Dr. Michelle Phoenix.

My curiosity for research began since I was an undergraduate student in the first cohort of the Child Health Specialization, Bachelor of Health Sciences (Honours) Program. I am thankful that the Child Health Specialization team continued to welcome me as part of the family even after I graduated. Thank you, Ms. Margaret Secord, Ms. Lorna Colli, and Dr. Stelios Georgiades, for your mentorship. Thank you, Alessia Greco, Mark Lee, Michael Wong, Joshua Koenig, and Christine Corso, for our continued work together to facilitate learning among students about child health.

I have learned that partnerships are critical in research in order to strive forward and have positive impact. Thank you to the McMaster Autism Research Team, Kids Brain Health Network, and CHILD-BRIGHT Network for our ongoing partnerships.

I am very grateful for funding support including: the Patient-oriented Research Award Fellowship – Transition to Leadership Stream – Phase 1 from the Canadian Institutes of Health Research (2020-2022), Graduate Fellowship in Patient-Oriented Research from the CHILD-BRIGHT Network (2020-2022), Conference, Training, and Travel Grant from the Cerebral Palsy Alliance Research Foundation (2019-2022), the Ontario Graduate Scholarship (annually from 2018-2020), and travel scholarship from the Kids Brain Health Network (2019).

I am very fortunate to have formed friendships during my PhD journey. I have learned and gained insights from my peers as we went navigated through the process of conducting qualitative and mixed methods research: Carrie Bullard, Karen Campbell, Shelby Deibert, Tessa Elliott, Paige Gehrke, Lin Li, Liz Orr, and Carly Whitmore. I am thankful for the friends that I have made in the School of Rehabilitation Science who continue to support me and encourage me to relax when I am in the midst of research activities: Silvana Choo, Young Heo, Sung Ha Kim, Isabel Rodriguez, Sirirat Bam Seng-Iad, and Joshua Yong. Thank you to my friends for all the times we spent in our graduate office, Mike Kalu and Janelle Gravesande.

Thank you to my friends since my days as an undergraduate student and who have been with me in every part of my PhD journey. Megan Schlorff, thank you for your words of kindness, providing a listening ear, and celebrating (big and small) highlights with me. Mathura Mahendren, you always reach out to check up on me and I know that your voice recordings of support and assurance will cheer up my day. Stanley Chen, thank you for always providing motivation, reminding me to trust the process, and encouraging me to believe in myself. As you like to quote Colin Kaepernick: “Don’t ask if your dreams are crazy, ask if they are crazy enough.”

Finally, thank you to my family who provided the most positive environment for me to complete my PhD journey. I know that it was not easy to transition having me work from home during the COVID-19 pandemic, and I am thankful that my family was willing to support me in this final stage. A special thank you to my niece, Jennifer To, for providing emotional support and being open to late night chats throughout my journey.

TABLE OF CONTENTS

LIST OF TABLES

CHAPTER ONE

CHAPTER TWO

CHAPTER FOUR

CHAPTER FIVE

CHAPTER SIX

CHAPTER SEVEN

CHAPTER EIGHT

LIST OF FIGURES

CHAPTER ONE

CHAPTER TWO

CHAPTER FOUR

CHAPTER FIVE

CHAPTER SIX

CHAPTER SEVEN

CHAPTER EIGHT

LIST OF SUPPLEMENTARY FILES

CHAPTER ONE

CHAPTER TWO

CHAPTER THREE

CHAPTER FOUR

CHAPTER EIGHT

LIST OF ABBREVIATIONS

BBD	Brain-Based Disability
BEST SIBS Study	BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS Study
CHILD-BRIGHT	Child Health Initiatives Limiting Disability Brain-Research Improving Growth and Health Trajectories
CIHR	Canadian Institutes of Health Research
ICF	International Classification of Functioning, Disability, and Health
iKT	Integrated Knowledge Translation
KT	Knowledge Translation
PFAC	Patient Family Advisory Council
POR	Patient-Oriented Research
PPEET	Public and Patient Engagement Evaluation Tool
RCT	Randomized Controlled Trial
SibYAC	Sibling Youth Advisory Council
SPOR	Strategy for Patient-Oriented Research
READYorNot™	Readiness in Youth for Transition Out of Pediatric Care
TD	Typically Developing
WHO	World Health Organization

DECLARATION OF ACADEMIC ACHIEVEMENT

This thesis is presented as a sandwich format, and is comprised of a general introduction, four independent manuscripts, and an overall discussion. At the time of preparing this thesis, the four independent manuscripts have either been published in peer-reviewed journals (Chapters 2, 3, and 5), submitted to a peer-reviewed journal (Chapter 4), or prepared for publication (Chapters 6 and 7). The candidate, Linda Nguyen, is the first author on all four of these manuscripts and was responsible for taking the lead in all stages of the research project including. The contributions of the candidate and co-authors for each manuscript is described below.

CHAPTER 2

Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Frontiers in Rehabilitation Sciences. 2021:52. doi: 10.3389/fresc.2021.724589

Contributions

Generated the research question: LN, SMJ, BDR, MK, JWG
Designed the study: LN, SMJ, BDR, MK, JWG
Extracted the data: LN
Analyzed and interpreted the data: LN, HD, SB, JH, SMJ, BDR, MK, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, HD, SB, JH, SMJ, BDR, MK, JWG

CHAPTER 3

Nguyen L, Bootsma J, Di Rezze B, Jack S, Ketelaar M, Gorter JW. Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: Protocol of a scoping review. BMJ Open. 2021;11(7):e053184. doi: 10.1136/bmjopen-2021-053184

Contributions

Generated the research question: LN, JB, SMJ, BDR, MK, JWG
Designed the study: LN, JB, SMJ, BDR, MK, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, JB, SMJ, BDR, MK, JWG

CHAPTER 4

Nguyen L, Bootsma J, Ketelaar M, Di Rezze B, Jack S, Gorter JW. Programs to prepare siblings for future roles to support their sibling with a neurodisability: A scoping review. Submitted to Current Developmental Disorders Reports.

Contributions

Generated the research question: LN, JB, SMJ, BDR, MK, JWG
Designed the study: LN, JB, SMJ, BDR, MK, JWG
Extracted the data: LN, JB

Analyzed and interpreted the data: LN, JB, MK, SMJ, BDR, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, JB, MK, SMJ, BDR, JWG

CHAPTER 5

Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Protocol of the BEST SIBS study: a qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition. Journal of Transition Medicine. 2021;3(1). doi: 10.1515/jtm-2021-0004

Contributions
Generated the research question: LN, SMJ, BDR, MK, JWG
Designed the study: LN, SMJ, BDR, MK, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, SMJ, BDR, MK, JWG

CHAPTER 6

Nguyen L, Jack SM, Davis H, Bellefeuille S, Arafeh D, Di Rezze B, Ketelaar M, Gorter JW. Roles of siblings of youth with a neurodisability during the transition to adulthood: A qualitative case study. To be submitted to a peer-reviewed journal focused on the health and development of children, adolescents, and young adults.

Contributions
Generated the research question: LN, SMJ, BDR, MK, JWG
Designed the study: LN, SMJ, HD, SB, DA, BDR, MK, JWG
Collected the data: LN
Analyzed and interpreted the data: LN, SMJ, HD, SB, DA, BDR, MK, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, SMJ, HD, SB, DA, BDR, MK, JWG

CHAPTER 7

Nguyen L, Jack SM, Davis H, Bellefeuille S, Arafeh D, Di Rezze B, Ketelaar M, Gorter JW. "Filling in the gap": A qualitative case study about the influence of family relationships on the identity formation of siblings of youth with a neurodisability. To be submitted to a peer-reviewed journal focused on age-specific issues and life transitions of youth.

Contributions
Generated the research question: LN, HD, SB, DA
Designed the study: LN, SMJ, HD, SB, DA, BDR, MK, JWG
Collected the data: LN
Analyzed and interpreted the data: LN, SMJ, HD, SB, DA, BDR, MK, JWG
Drafted the manuscript: LN
Review and provided feedback of the manuscript: LN, SMJ, HD, SB, DA, BDR, MK, JWG

CHAPTER ONE

Introduction

An overview

As children and youth with a chronic health conditions (CHC), including developmental disabilities and neurodisabilities, age, they will be required to transfer from the pediatric setting to the adult health care system in Canada. A non-categorical approach is used in this dissertation to describe children and youth with a CHC, which is defined as an approach where the focus is on the common experiences and challenges based on general health characteristics instead of a specific CHC (1). During the transition to adulthood, adolescents experience changes including biological, emotional, and behavioural changes (2). These changes lead to the development of their behavioural functions, such as self-control, judgment, planning, and problem-solving (3). During this period of time, a developmental task for adolescents is to become more independent in activities and roles in life (3).

Youth with CHCs have the additional pressing task of learning how to take care of their health. In the universal public health care system in Canada, children and youth with a CHC (ages 0-18 years old) typically receive health care services in the pediatric healthcare system, such as children's rehabilitation centres and children's hospitals (4). Youth and families will have to prepare for the transition process, in which they will have to transfer from receiving healthcare services from pediatric to adult systems of care that typically occurs at age 18 in Ontario, Canada (5). Youth often experience challenges during healthcare transition, because they need to learn how to navigate the unfamiliar adult healthcare system. Families, including siblings, can provide support to youth during healthcare transition. These supports can include emotional support, transportation, reminders to take medications and attend healthcare appointments, providing advice for general information and health information (6,7).

Typically developing (TD) siblings have a unique relationship to their sibling with a CHC as they often grow up in a shared home environment, in which they know each other's life, preferences, and healthcare history (8,9). Previous research has focused on the general descriptions of roles that TD siblings might have to their sibling with a neurodisability, such as being a role model, mentor, confidant, friend, or caregiver (8,10).

A current knowledge gap is in the understanding the role(s) and responsibilities of TD siblings during the healthcare transition of their sibling with a CHC, as well as the factors of the sibling relationship that could influence the roles of TD siblings (10). In 2017, a needs assessment survey was conducted with 260 responses completed from siblings of individuals with intellectual disabilities in Ontario, Canada (11). Survey responses identified that siblings prioritized tools, resources and information to support their sibling with an intellectual disability on topics, such as finances, funding, and mental health supports for the whole family (11). There is a need to synthesize current resources in Canada to support siblings in their role(s) in the healthcare management of their sibling with a CHC. To address this knowledge gap and need, collaborations with relevant stakeholders are critical. Stakeholders have a right to be involved in research that is about them. The expression of "nothing about us without us" was first used by disability rights activists to advocate for the full participation of stakeholders in policies that concern them (12). This expression has also been applied by stakeholders who wish to partner in healthcare initiatives and research that is about them (13–15). A patient-oriented research approach can be used to engage with patients, who are broadly defined as any individual with a personal experience of a health issues and their family members (16). Patients can be considered as stakeholders, who have experiences that can help to inform the relevance of research, provide new insights to better understand the research findings, and ensure that the research findings can be shared meaningfully with the community (16).

To inform all steps of the research process undertaken for this project as well as to address issues important to siblings and families, I used a patient-oriented research approach (17) and established the Sibling Youth Advisory Council (SibYAC) in 2018, in the early stages of this work. From 2018-2022, there has been a range in membership from two to six adult siblings (ages 21-28 years old) of a sibling with a disability living in Alberta and Ontario, Canada.

The overall objective of this dissertation was to develop an understanding about the experiences of Canadian siblings of youth with disabilities during the transition to adulthood, and to synthesize current resources and programs to support them. The following sections outline the definitions of key terms and concepts in this dissertation. A literature review is also provided about the relationships of siblings with and without a CHC, including key characteristics and developmental changes. Then, the philosophical, methodological, and theoretical understandings that informed this dissertation is described. This also includes a description about my position as a researcher, and how my values, beliefs and experiences shaped the research process including my partnership with the SibYAC. As this dissertation consists of a collection of manuscripts that have been submitted and published, and this chapter concludes with a brief summary of subsequent chapters. The manuscripts include a review and qualitative document analysis to synthesize resources for siblings of youth with chronic health conditions (Chapter 2), a protocol and results of a scoping review about programs to prepare siblings in their future roles to their sibling with a neurodisability (Chapters 3 and 4), protocol and results of a qualitative study to understand the experiences of siblings of youth with a neurodisability during the transition to adulthood (Chapters 5, 6, and 7). Details about each manuscript is provided at the end of this chapter.

Population of youth with a neurodisability

In North America, approximately 15% to 18% of youth (ages 0 to 17 years old) have a chronic health condition (CHC) (18,19). Chronic health conditions include physical or mental illnesses and developmental disabilities (20). Within developmental disabilities, congenital and acquired disabilities are included, and more specifically, neurodevelopmental disability or neurodisability. Recent statistics identified that the prevalence of neurodisability is one in eleven between the ages of 4-11 years old based on data from the National Longitudinal Survey of Children and Youth in Canada and a health database in British Columbia (21). Neurodisability can be broadly defined as a group of congenital or acquired long-term conditions due to an impairment of the brain and/or neuromuscular system that may create functional limitations (22). Neurodisability diagnoses can be categorized into: 1) developmental that describes functional characteristics related to child development, such as autism spectrum disorder or attention-deficit and hyperactivity disorder or 2) medical based on a primary underlying etiology such as Down syndrome or fetal alcohol spectrum disorder (23). Recent shifts in pediatric healthcare have focused on providing care and supports to children with a CHC and their family using a noncategorical approach to focus on the common experiences of health conditions instead of focusing on a single health condition (1). Considering the wide spectrum and heterogeneity of neurodisabilities, this non-categorical approach can help to understand the comprehensiveness of care and supports to address the varying needs of the individual and family (1,23,24).

For the purpose of this chapter, the general term of ‘disabilities’ will be used using person-first language. This dissertation focuses on disabilities from a rehabilitation science and health perspective with the recognition that there is ongoing debate and dialogue about the language use and the paradigm for service provision with children and youth with disabilities (25). Pediatric rehabilitation science has experienced a shift from a traditional biomedical approach (i.e. ‘fixing’ of the impairments) to a focus on the goals of children and adolescents that best aligns with their abilities and wishes (25). For example, occupational therapists have advocated for the adoption of a critical approach to work with individuals with disabilities. Instead of asking the questions of what an individual with a disability cannot do, health care professionals including occupational therapists can ask about what can be learned from an individual with a disability (26). Throughout the process of completing my dissertation, I have ongoing conversations with the SibYAC and participants about their preference in how they refer to disability. In my writing and knowledge translation activities, I include a preface that I use person-first language with the recognition that every individual has their preference. I am also aware about continuing to create a safe and welcoming space to ask the SibYAC and participants about their preference in discussions about disability and services.

Framework describing factors impacting the development of youth with a neurodisability

Multiple factors can affect the development of youth with a neurodisability, and these factors can relate to the individual youth such as personal strengths and needs, availability of social supports, and the settings that the youth interact with (3). An understanding of how these factors can facilitate or hinder the development of youth with a neurodisability can be viewed using the International Classification of Functioning, Disability and Health (ICF) framework. The ICF framework was developed in 2001 by the World Health Organization as a biopsychosocial model to classify the factors that influence the health and wellbeing of an individual (Figure 1) (27). The ICF is comprised of five domains:

- i) body functions and structure, defined as the body's physiological functions of body systems and anatomical parts of the body;
- ii) activities, defined as the execution of a task or action;
- iii) participation, defined as involvement in a life situation;
- iv) environmental factors, defined as the physical, social, and attitudinal environments; and
- v) personal factors, defined as the background of an individual that is not part of their health condition, such as gender, race, or age (WHO, 2001, ibid. p. 9, 15)

The ICF framework provides a broad perspective towards optimizing the health of children. As children grow up, their health and functioning can be viewed in the context of the family system (27). The goals of healthcare services and programs, including those that address healthcare transition, can consider how to support the youth with disability, as well as the whole family.

Children and youth grow up in varying family structures. In a Canadian census that examined the household of children aged 0 to 14, children may live in households with both parents or one parent who may be their biological, step, or adoptive parents (28). Some children may be living with their grandparents, other parents, or in foster care (28). In this dissertation, the focus is on families in which the primary caregiver(s) is the parent, with or without support from extended family, with more than one child in the home setting.

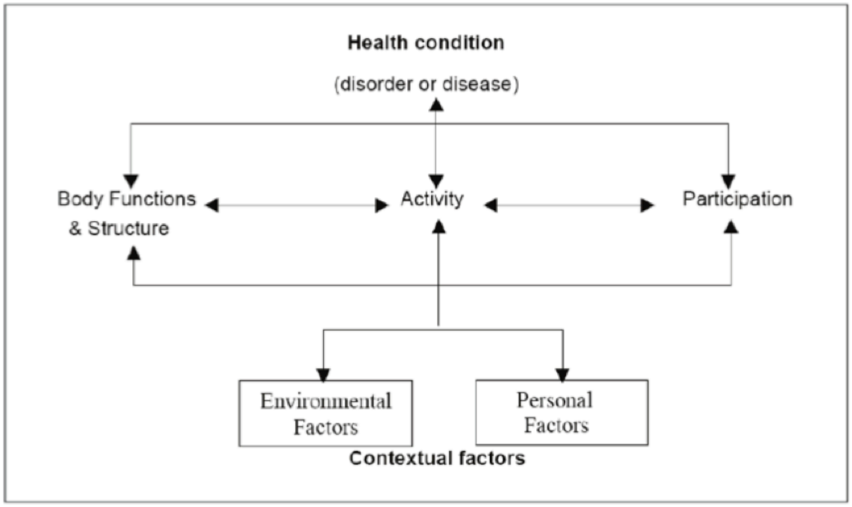

Figure 1. *The International Classification of Functioning, Disability and Health* (27).

Youth challenges during transition to adulthood

Youth are experiencing biological, social, and emotional challenges during the transition to adulthood, or the period between the start of puberty and early adolescence (approximately 10 years of age) and through middle and late adolescence (until 18 years of age) (2). Emerging adulthood is a developmental period (18 to 25 years), where youth are transitioning from adolescence to adulthood (29). During emerging adulthood, youth often have multiple opportunities to explore their interests and goals related to school, work, family, and leisure (3,4,29–31). Youth will have to learn how to navigate new environments, including healthcare, education, recreation, and social services (3). However, during this developmental period, youth with a disability may experience distinct barriers that their peers without a disability do not face (4). These barriers may relate to personal factors, such as low expectations or physical limitations, or to the environment, such as lack of accessibility in buildings and availability of health services (4,32). Youth with a disability have reported challenges related to attending postsecondary education, securing employment, developing long-term relationships, and navigating adult care services (29,33). Youth with disabilities will need to learn how to overcome barriers as they navigate new environments during their transition to adulthood. When youth with disabilities are supported to overcome these barriers in their environment, the appropriate fit that they have with their environment can lead to positive outcomes on their health and development (34).

Healthcare transition for youth with disabilities

One of the critical transitions that youth with disabilities will experience is the transition from pediatric to adult healthcare. Healthcare transition is a process that is defined as the "purposeful, planned movement of adolescents with chronic medical conditions from child-centred to adult-oriented healthcare" (10, p.786). An element of this transition process is healthcare transfer, a one-time event when the youth leaves the pediatric health system and enters into the adult health system (35). Youth and families often experience multiple challenges during transition because they will need to leave the pediatric system that they have known and they will have to learn how to navigate the new adult healthcare system (4). With the numerous challenges that youth and families often face in the new adult healthcare system, they often describe their experience with healthcare transition as "falling off a cliff' (4). Many health concerns during childhood will continue into adulthood, along with additional health issues that may arise (36). The lack of a successful healthcare transition contributes to increased emergency room visits, increased length of hospital stays, frequent access of healthcare services that leads to increases in overall healthcare costs with less than optimal use of healthcare services (37–39).

The process of transition is encouraged to start early with discussions with the youth, their family, and healthcare professionals in order to prepare for the healthcare transfer (40,41). These discussions might address topics such as specific roles of family members, anticipated challenges, and access to additional resources and information to prepare for transition (42). Consensus statements, published in 2002, from the American Academy of Pediatrics, the American Academy of Family Physicians, and the American College of Physicians-American Society of Internal Medicine stated the importance of providing the necessary resources to support and facilitate transition of youth with disabilities into adulthood (43). The recent consensus statement from the Canadian Pediatric Society in 2022 further outlines and emphasizes support to be provided before and after transition to adult care (44). This support includes applying a stepwise approach to increase the independence of youth in the management of their healthcare, and providing a comprehensive summary about health transfer that includes the needs of the youth and families (44). After the transfer to adult care, the support include making sure that youth attend appointments, involve the family based on the wishes of the youth, and regularly measure health outcomes (44). Therefore, the transition process can be a collaboration with youth, families, and healthcare professionals.

Family support

In a family, parents are typically the primary caregivers of children and youth with neurodisabilities. They are responsible for coordinating care and services for their children, and they face the challenge of supporting their children and while caring for themselves as they age (45–47). Grandparents may also offer support to the family. A scoping review conducted in 2020 identified that grandparents often provided emotional support, such as providing encouragement to their adult children and enjoying emotional closeness with their grandchildren; financial support to assist the family; or instrumental support such as providing childcare or assisting with transportation (48).

Youth can refer to their family for support to be prepared and ready for healthcare transfer. In Ontario, Canada, this transfer typically occurs at the age of 18. Pediatric healthcare is often family-centred with shared decision-making with the child and parents (49). Principles of family-centred practice emphasize the partnership between parents and healthcare professionals (50–52). While the child is growing up, the parents are often considered as the 'experts' who are most likely to have been involved throughout the care of their child; they know their child best including the child's strengths, areas of improvement, and goals (53). When youth are preparing for transition, they may begin to learn how to become independent in the self-management of their healthcare (6). This transition process can begin gradually in a stepwise approach, when the roles of the youth, parents, and healthcare professionals begin to change (44). The youth, parents, and healthcare professionals can be considered as a part of a partnership alliance, known as a shared management model (54). During the early stages of the transition process, parents may start to move from a management role to the role of a supervisor; for example, making sure that their child takes their medications and attend healthcare appointments, and teaching their child to ask questions to their healthcare professionals (6,42,55). Parents may then gradually shift towards the role of being a consultant, who can provide support; for example, parents can provide general and healthcare advice (6,42,54–57). Healthcare professionals can have the role of being an enabler and collaborator by facilitating a gradual approach to encourage the youth to be independent; for example, by providing the date of the next healthcare appointment directly to the youth (42). Parents also require support during the healthcare transition of their child, as they may feel anxious about possible risks as their child increasingly takes on more responsibilities (6). Healthcare professionals can offer support to parents, such as explaining the rationale for the change in roles during healthcare transition and teaching coping skills (6). Overall, healthcare transition is a gradual process in which there will be different ways for how parents and healthcare professionals can support the youth to take charge of their health.

Sibling relationships in families of youth with disabilities

Context of sibling relationships in Canada

There are varying types of sibling relationships including: biological siblings, in which siblings have the same parents; half-siblings in which siblings have one common biological parent; and step-siblings who are not genetically related to each other but their parents are in a relationship (58). The Canadian census data from 2016 identified there are approximately 31% of couples who have two or more children and approximately 39% of lone-parents with two or more children (59). There are approximately 2-4% of children who are living with half- or step-sibling (59). However, there is a lack of data in Canada specifically about children with disabilities with the last census data that collected this information in 2006 (60,61). To address this gap, recommendations have been provided to monitor and track the needs of children with disabilities and their families (62). Research has identified the nature of how development of the sibling relationship over time and the factors that influence this relationship as described in the following section.

Development of sibling relationship over time

Each sibling relationship is unique, and the nature and quality of the relationship may evolve through the different developmental periods. Typically developing (TD) siblings will have changing perceptions of their relationship to their sibling with a disability as they grow up together and are a part of each other's life journey as ultimate fellow travelers (63,64).

Childhood to adolescence. During childhood and adolescence, siblings often live in a shared home environment (8). When siblings grow up together, they are often close in which they share and learn about the details about each other's life including their relationships and history of their health (9). Siblings have also described that their life at home is often "chaotic" and a "struggle" when one sibling has a disability (65). At a young age, siblings often become aware that they need to support their family in ways that are different from their peers (66,67). For example, they may recognize the important role that they have to care for their sibling and cancel their own plans when their parents were tired or there was a family situation where they had to stay at home with their sibling (66). During day-to-day life as children, siblings can have roles such as being a role model, mentor, confidant, or friend to their sibling with a disability (8).

Adolescence to emerging adulthood. During this time, emerging adults in general have identified that they experienced more emotional exchanges with their siblings, such as warmth (68). There is often less conflict and rivalry compared to childhood as the emerging adult siblings have developed a more mature perception and understanding of their sibling relationship (29,68). When siblings transition to emerging adulthood, they will also experience changes in their lives such as moving away from home for employment, post-secondary education, or personal reasons (63). Other changes could occur in the sibling relationships as the siblings start to build a life outside of the intermediate family home with new experiences and relationships with people (63). For example, siblings might start their own family and start to raise their own children, so they assume a new role and set of responsibilities (63).

The changing dynamic of sibling relationships can vary and some previous research has considered this dynamic as a U-shaped curve of closeness over a period of time (69). The highest and closest relationship between siblings often occur during childhood, followed by a decrease in closeness during adolescence and emerging adulthood, followed by an increase in closeness later in adulthood (See Figure 2, 66). A similar U-shaped curve can describe the amount of communication between siblings, which includes in-person contact, phone calls or texts (69).

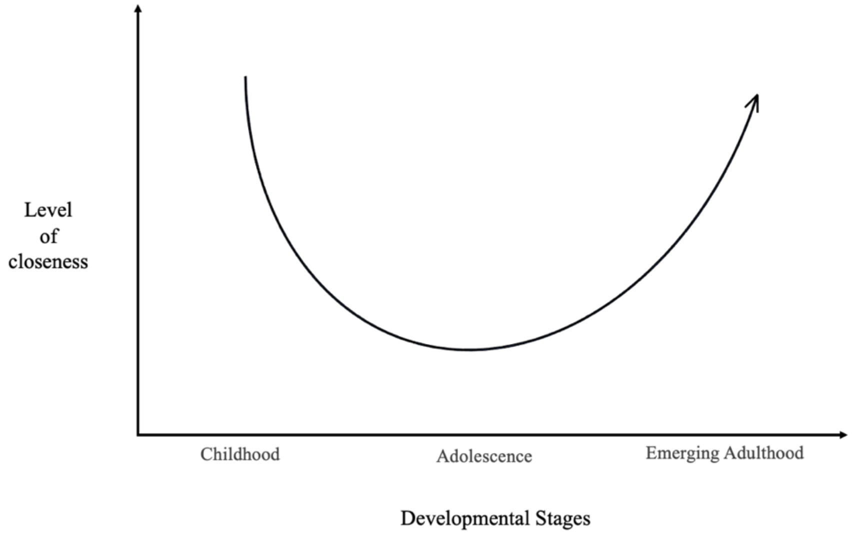

Figure 2. A visual depiction to describe the changes in the level of closeness between siblings during the developmental stages of childhood, adolescence, and emerging adulthood.

Influential factors on the sibling relationship

There are multiple factors that influence the development of the sibling relationship over time, and the most commonly described factors in the literature includes personal characteristics, gender, and birth order of the siblings (70).

Personal characteristics. The temperament of siblings, both the TD sibling and the sibling with a disability, can affect the sibling relationship. Temperament can be defined as the behavior style of individuals in relation to others and the environment (71), which can be characterized by activity level (i.e., movement and high energy) and adaptability (i.e., the tendency to approach new individuals and situations) (72). Difficult temperaments can be defined as having high activity level and low adaptability (71). In a study by Brody and

Stoneman (1993), siblings with positive relationship occur when one child has a low activity level and the other child has high adaptability, whereas the most conflicts would occur when one child has a high activity level and the other child has low adaptability (71). Temperament can include other dimensions, such as mood (e.g., amount of friendly behaviour compared to unfriendly behaviour) and persistence to pursue an activity (73). Additional studies have shown how temperament can be a moderator of how siblings manage their stresses and conflicts within the family (74–76). The temperament of TD sibling and sibling with a disability can affect the level of conflict experienced in the relationship (77).

Health condition. The type of disability and functioning of the sibling can have an effect of the sibling relationship. In the literature, there is mixed research about the feelings that TD siblings have towards their sibling with different types of disabilities; some studies identified how siblings of individuals with autism spectrum disorder experienced less frequent contact (78,79), while other studies described that the level of warmth experienced by TD siblings of siblings with intellectual disabilities varied (80–82). Alongside the health condition, the behaviour of the sibling with a disability can further influence the level of closeness in the relationship. Some TD siblings described that the difficult behaviours of their sibling with autism spectrum disorder, such as aggression or not wanting to speak with others, affected the quality of the relationship (83,84). Over time, some TD siblings adjusted and came to understand the behaviour of their siblings which led to a positive viewpoint of the sibling relationship (83). The level of care that is required for the sibling with a disability also affects the sibling relationship, in which some TD siblings made decisions about whether to provide support with daily activities, including personal care and hygiene such as showering, toileting, and feeding (85,86).

Gender. The gender of the TD sibling and sibling with a disability can affect the development of their relationship, and the roles that TD siblings might choose to have to their sibling with a disability. Females are perceived to be more involved in the relationship with their sibling. Sibling dyads of the same gender often have the closest bonds (68,87–92), with female-female dyads described to have the most intimate and warm bond compared to other sibling dyads (68,87,88). These findings about the influence of gender are also applicable to relationships between TD siblings (68).

Birth order. The birth order between the TD sibling and sibling with a disability can affect the experiences that they have in the relationship. Older siblings may take on roles of authority and responsibility to their younger siblings (93,94). For example, older siblings might act as sources of support who the younger siblings might turn to for advice regarding social and academic problems (95).

Emotional impact of the sibling relationship on typically developing siblings

Both the TD sibling and sibling with a disability are growing up and will have an impact on each other. Empirical studies have focused on the emotions and effects of having a sibling with a disability on the TD sibling (63,96). The TD siblings may experience a range of emotions on a spectrum that includes joy when they can spend time with their sibling with a disability; guilt with how they treat their sibling with a disability, including how they might have treated their sibling in the past; resentment when their sibling with a disability demands increased parental attention, or stress and frustration about care services for their sibling with a disability (81). There is a mixture of effects that TD siblings experience as an outcome from having a sibling with a disability; positive effects might include having greater empathy, compassion and understanding (96–98), while negative effects might include having depression or anxiety (96,99). Taking a lifespan approach, the emotions and effects felt by TD siblings can vary and change as they grow up. The TD siblings often have both positive and negative experiences in their relationship to their sibling with a disability at different points in time. Throughout these experiences, TD siblings wanted to establish and maintain some normalcy within the family (65).

Roles and responsibilities of siblings

Siblings may have different roles and responsibilities in multiple processes that occur during the transition to adulthood of their sibling with a disability, for example, the transition from pediatric to adult healthcare or the transition from high school into postsecondary education and/or employment (100,101). During healthcare transition, the youth with a disability and family need to be prepared for the transfer to adult care services. Youth with disabilities may refer to their family, including their siblings, for support.

Support can have many forms including:

i) concrete or tangible, support such as providing assistance;
ii) emotional support, such as listening, empathizing, or providing comfort;
iii) advice support, such as providing suggestions or information;
iv) or esteem support, such as expressing confidence or encouragement (102,103).

Siblings can provide support in formal or informal roles to their sibling with a disability based on the family circumstances, and sometimes, siblings are assumed to have these roles by the family (10). There are a variety of roles that siblings might have, which can change over time. The roles of TD siblings may include a mentor, role model, caregiver, advocate, legal representative, informal service coordinator, or friend to their brother or sister with a disability (10,104). Many of these roles apply to not only healthcare transition, but the broader transition to adulthood for the sibling with a disability.

All members of the family, including TD siblings, need to be prepared for both healthcare transition and the transition to adulthood for the sibling with a disability. The TD siblings can have the autonomy to choose whether to take on different types of roles, and they need to be prepared and empowered to take on certain roles. While TD siblings might have specific roles to their sibling with a neurodisability, there is limited understanding about the experiences that they have in these roles and how they are being supported.

Research gap on siblings of youth with a neurodisability

First, there is a current knowledge gap about how TD siblings decide on their role(s) and responsibilities during healthcare transition, as well as whether mediating factors of the sibling relationship (e.g., personal characteristics, health condition, gender, birth order) influence this process. Siblings are not always involved in discussions about the planning of their sibling with a disability (105,106). Some families have discussions about the roles that siblings could have, but often there are no formalized plans (107). There is a need to generate knowledge in order to have a better understanding of how TD siblings come to assume current roles and preparing for future roles, specifically when their sibling with a disability is transitioning to adulthood.

Second, there is a gap in our understanding about existing programs and resources that are available for siblings to prepare them in their roles with healthcare management to their sibling with a disability. In Canada and the United States, there are Sibshops (ages 8 to 13 years old) and Sibteen sessions (ages 13 to 17 years old) that provide an opportunity for siblings of individuals with disabilities to connect and share experiences with each other (108,109). However, these types of programs are mainly focused on supporting the siblings but does not focus on the preparation of roles that siblings might have such as with healthcare management. Siblings might choose that they want to have certain roles to their sibling with a disability, but they need to identify supports and resources for these roles (110). A Canadian Siblings Needs Assessment Survey was conducted in 2018 that collected responses from 360 adults (≥20 years old) with 87.2% of respondents in Ontario who had a sibling with a disability. Adult siblings mostly commonly identified concerns related to housing and finances to support their sibling with a disability (11,111). Siblings often require greater support and resources if they choose to have as a future caregiver to their sibling with a disability, but the age and readiness of the siblings to step into this role needs to be considered when providing resources (112). Adult siblings want specific supports, such as how to navigate services in the adult system, improved delivery of systems in general, and additional funding and respite care (9,106). Adolescent and emerging adult siblings also need to be prepared and supported in their roles, but there is currently limited information available for this age group of siblings. There is a need to synthesize knowledge about current programs that provide information to prepare TD siblings in the roles that they choose, so that they can then in turn support their sibling with a disability during the transition to adulthood, including the transition from pediatric to adult healthcare.

Philosophical and methodological underpinnings of this dissertation

In order to synthesize existing knowledge and generate new knowledge to address how siblings experience and prepare for their roles to their sibling with a disability, an understanding of the philosophical assumptions that underpin this dissertation is needed. Philosophical assumptions shapes how knowledge in this dissertation was generated, the nature of the relationship between myself as a researcher and the research, and the methods applied in this dissertation. There are four key philosophical assumptions that inform research practices (113):

1) ontology, refers to the beliefs about the nature of reality;
2) epistemology, describes how knowledge is known and the assumptions about how the relationship between the researcher and participants is understood;
3) axiology, reports the values brought into the study by researchers, including how researchers positions themselves within the context and setting of their research;
4) methodology, characterized by the principles to conduct research.

These philosophical assumptions can be applied within interpretive frameworks to conduct research (113,114). The interpretive framework that was used to inform this dissertation is social constructivist, also described as interpretivism (114,115). Table 1 describes how each philosophical assumption aligns with social constructivism that was used to guide this dissertation. Using a social constructivist paradigm, research aims to understand the meaning that individuals have about their experiences, and these experiences are formed through interactions with others as well as through historical and cultural norms (113). Researchers aims to address the processes of interactions between individuals within specific settings. Among researchers, there is recognition that their own background influences their interpretation of the data. To conduct the scholarly works in this dissertation, I needed to understand how I position myself as a researcher, as well as how my personal and professional experiences shaped this dissertation.

Table 1. Social constructivism and associated philosophical beliefs to guide this dissertation.

	Ontological beliefs	Epistemological beliefs	Axiological beliefs	Methodological beliefs
Social constructivism	There are multiple realities, constructed through lived experiences and interactions with others within cultural, historical, and social contexts.	Reality is co-constructed between the researcher and those being researched, as well as shaped and interpreted by individual experiences.	Values are acknowledged and negotiated between individuals.	Research findings are generated inductively, with data that is obtained using techniques such as interviews, observations, and texts.

Adapted from Creswell & Poth (2013).

Researcher positionality: Who I am and how I position myself within this work

Throughout my PhD journey, I have reflected on 'who' I am along with the 'how' and 'why' I am embarking on this journey to conduct this work. As a person, I am the daughter of immigrant parents from Vietnam, a sister (with an 18-year age gap between myself and my older sister), and an aunt (with a 7-year age gap between myself and my niece). My experiences of growing up in a multigenerational family with different roles motivated me to further understand unique sibling relationships that are influenced within the family environment and societal contexts. My personal values stem from both the Western culture in Canada and the Vietnamese culture from Asia. In the Vietnamese and Asian cultures, there is a collectivist approach in which everyone in a family to care for each other (116). I recognize the importance of multiple family members coming together to care for each other. I live in one household with my father, mother, older sister, brother-in-law, niece. When there are situations, each family member is responsible to take care of each other. An example is around babysitting, as my older sister watched over me when was a young child and in turn, I was also responsible to pick up and care for my niece when she was young. This babysitting role was an unpaid position that was an inherent responsibility within the family. I recognize the importance of adopting a collectivist approach to care for the whole family.

In parallel to my values of collectivism, I also adopt the values of individualism from the Western culture in Canada. As my parents are aging and are in retirement, I recognize that I have a responsibility along with the whole family to take care of my parents. This level of care is an ongoing conversation with all members of my immediate family that includes a balance with personal interests that I would like to pursue. Some care tasks that I carry out for my parents, are similar to the role of a young carer. A young carer is defined as a child or youth between the ages of 5 to 18 years old in a caregiving role for a family member with a health condition, disability, addiction, mental illness, or language barrier (117). My main role is to make sure that my parents have access to supports and services that they require. Since I was about 8 years old, I translated for my parents when they accessed services, including the healthcare system. Sometimes, the responsibility to be a translator included missing school to be present with my parents at appointments. As I grew older, I identified that there is a collective responsibility of the whole family to support each other, and I learned how to advocate for myself by communicating about my priorities with the family. The diffusion of responsibilities within the family is dynamic and can continually change based on different priorities from each family member. For this reason, I believe that ongoing family conversations are very important.

My overarching values of collectivism and individualism influences how I approach my research, and my personal experiences further influence how I view the topic of this dissertation about healthcare transition. I have experienced the challenges in learning how to navigate the healthcare system. Some challenges that youth with disabilities and their families have experienced, such as communicating with healthcare providers, are experiences that resonate with me. I developed skills that youth also had to develop when they were preparing for healthcare transition. Tools can help youth to develop skills to communicate their needs with healthcare providers; for example, the MyTransition App (118) includes the MyHealth 3-Sentence Summary in which youth create statements to include information about their: 1) age, medical condition, and brief medical history; 2) treatment plan; and 3) questions or concerns to discuss during the health visit. Similar to youth, I also had to develop these communication skills when discussing the medical history and treatment plan of my parents during their healthcare appointments, and I had to be able to clearly translate between my parents and their healthcare providers. Not only was I communicating with my parents, but I was also providing updates to my older sister. Overall, I view the healthcare transition is a collective experience with the whole family, and ongoing family conversation is critical to ensure that all members are informed.

As a scholar, my academic journey was influenced by my educational background. I graduated from the Bachelor of Health Sciences (Honours) Program, Child Health Specialization in 2015. Throughout my undergraduate studies, I often wanted to ask the “why” questions, and I enjoyed reading research papers to see what other questions and knowledge we can generate. In 2017, I completed my Masters in Rehabilitation Science under the supervision of Dr. Jan Willem Gorter. For my Master’s thesis, I conducted a qualitative study to better understand the effects of botulinum toxin treatment in children and adolescents with cerebral palsy from the perspectives of parents. Since conducting the qualitative study, I recognized the value of understanding families’ perspectives in research. For this reason, I wanted to consider how I can continue to connect with families during my PhD studies. I was motivated to continue with graduate studies and decided to pursue a PhD in Rehabilitation Science to enhance the care with and for families of children and youth with disabilities.

With my interest to gain knowledge and experience to engage with partners in research, I was able to develop foundational knowledge about patient and family engagement through the Family Engagement in Research Certificate of Completion Program hosted by McMaster University Centre for Continuing Education, CanChild Centre for Childhood Disability Research, and Kids Brain Health Network (119). I also had the opportunity to work as a graduate research assistant (Sept 2017 – Dec 2021) with my supervisor, Dr. Gorter, as a Co-Principal Investigator for the CHILD-BRIGHT: Child Health Initiatives Limiting Disability – Brain Research Improving Growth and Health Trajectories, with the project titled, READiness in Youth fOR traNsition Out of pediaTric Care ('READYorNot' Brain-Based Disabilities) funded by the Canadian Institutes of Health Research – Strategy for Patient-Oriented Research (120). As a graduate research assistant, I had the opportunity to learn how the project partnered with the Patient and Family Advisory Council (PFAC). Supplementary File 1 provides details about my learnings from these experiences, which I applied to my partnership with the SibYAC.

Theoretical underpinnings

Theories can be helpful to guide the research process, in which theories can provide definitions about key constructs that can be used to inform the research problem or phenomenon being studied (121). The following theories were used to inform the design and conduct of the studies in this dissertation.

Transition theory (122)*:* The transition theory was developed by Meleis and colleagues to describe the different types of transition, properties of the transition experience, and how individuals experience the process of transition that leads to specific outcomes (122).

Transition is a complex process that can be characterized by the following essential properties:

i. Awareness: Related to perception, knowledge, and recognition of the transition experience from an individual;
ii. Engagement: Degree of involvement from an individual during the transition process;
iii. Change and difference: Transitions can be a result *of* change (e.g., due to an event) and *in change* (e.g., occurrences during the transition process)*;*
iv. Time span: Transition is characterized by a time period with an eventual endpoint;
v. Critical points and events: Critical points or marker events can involve increasing awareness of change or active engagement in the transition process.

This theory can inform how TD siblings experience the process of transition from pediatric to adult healthcare and overall transition to adulthood of their sibling with a disability.

Bioecological systems theory. This theory was developed by Urie Bronfenbrenner, a developmental psychologist, to describe person-environment transactions. Bronfenbrenner (1979) proposes four ecological systems: *the microsystem, the mesosystem, the exosystem, and the macrosystem*. The *microsystem* encompasses the individuals within the immediate environment of the TD system including parents, siblings, and healthcare providers. The *mesosystem* encompasses the relationships that are developed based on the interactions between the microsystems such as the relationships between TD siblings, sibling with a disability, parents, family members, and healthcare providers. The *exosystem* is the environment that impacts but does not directly involve the TD siblings such as the relationship between parents. The *macrosystem* is comprised of external systems that influence the whole society that TD siblings live in, such as healthcare, school, community, and policies. Bronfenbrenner (1986) later proposed the *chronosystem* that describes the influence of environmental changes on an individual over time. These changes may include developmental transitions, such as adolescence and/or young adulthood, and events, such as having a chronic illness. These ecological systems are dynamic and interact with each other. There could be changes within the sibling relationship that is influenced by environmental factors, including the family context and external systems. Figure 3 provides an adaptation of the ecological systems that influence the development of TD siblings and subsequently relationship with their sibling with a disability. This dissertation focused on the sibling within the microsystem (e.g., sibling with a neurodisability, other TD siblings, and parents/caregivers), and the mesosystem (e.g., their relationships with their sibling, other TD siblings, and parents/caregivers.

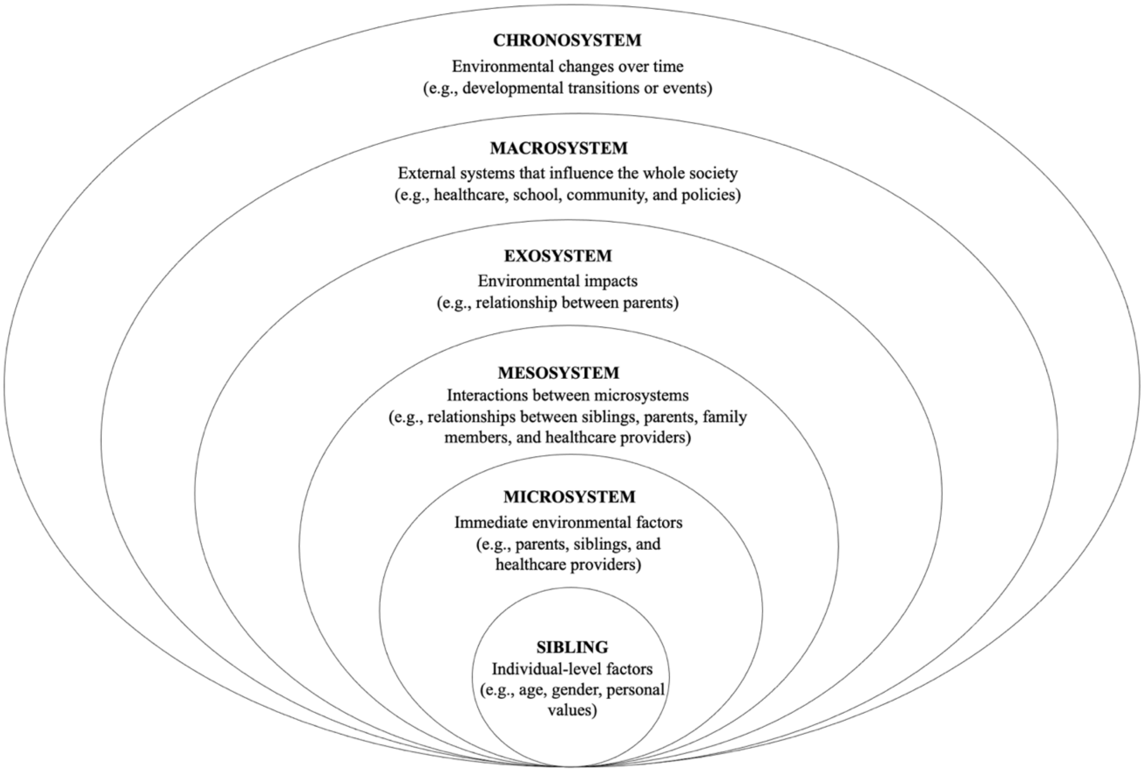

Figure 3. *The ecological systems that influence the development of typically developing siblings, and their relationship with their sibling with a disability. Adapted from Bronfenbrenner (1979, 1986).*

Patient-oriented research

Patient-oriented research (POR) is increasingly being recognized as an important standard and practice in applied health research. According to the Canadian Institutes of Health Research (CIHR), POR is defined as "a continuum of research that engages patients as partners, focuses on patient-identified priorities and improves patient outcomes. This research, conducted by multidisciplinary teams in partnership with relevant stakeholders, aims to apply the knowledge generated to improve healthcare systems and practices" (17). The term 'patient' can be broadly defined to include patients with health conditions, and their families and friends (17).

Engagement of patient stakeholders in research can have impact at different levels, including for researchers, patient stakeholders, and the research. The research impact might include increased relevance of the research topic to the target population (123,124), use of appropriate recruitment materials (125), access to hard-to-reach populations (123,124), development of user-friendly products (124,125), and increased dissemination and mobilization of study findings that is meaningful to the community (126,127). Patient stakeholders have identified benefits of their engagement in research, including development of new knowledge and skills (128–131), opportunities to be a part of a social network (128,131), and an increase in confidence and self-esteem (127–129,132). Reported benefits on researchers during partnerships with patient stakeholders include increased understanding of the perspectives of patient stakeholders, development of rapport with the community, increased motivation, and increased satisfaction (127,128,133,134). Overall, partnerships with stakeholders in research can have mutual benefits on not only the research itself, but also on researchers and stakeholder partners (135,136).

While there is increasing recognition about the value of POR, several systematic reviews have reported on the challenges in POR practices (125,126,137). Among these systematic reviews, a commonly cited challenge among research studies is logistics to needing extra time to complete research, flexibility with the process and direction of research, time constraints from patient stakeholders and researchers, and additional funding required (125,126,137). Logistic challenges experienced by research teams can lead to feelings of frustration and thus lack of meaningful engagement (125,126,137). There is also a concern that engagement of patient stakeholders could be a form of tokenism, defined as the "difference between…the empty ritual of participation and having the real power needed to affect the outcome" (138). Careful considerations can address how patient stakeholders can be engaged meaningfully in research.

Different frameworks and key principles can be used to identify strategies to build and sustain partnerships with patient stakeholders (139). The Strategy for Patient-Oriented Research outlines a Patient Engagement Framework (17) with guiding principles to support partnerships, and a description of these principles is provided in Table 2.

Table 2. A description of principles in the Patient Engagement Framework in the Strategy for Patient-Oriented Research from the Canadian Institutes of Health Research (17).

Principle	**Description**
Inclusiveness	To integrate diverse perspectives of patient stakeholders that reflect their contributions in research.
Support	To provide flexibility to ensure that patient stakeholders can fully contribute to discussions and decisions in safe environments that promote honest interactions, cultural competence, training, education.
Mutual respect	To create an environment in which all members of the research team acknowledge and value each other's expertise and lived experiences.
Co-build	To share opportunities for all members of the research team to work together from the beginning of the project, including identifying problems and gaps, set priorities, and produce and implement solutions.

Similar principles have been identified in the literature (127,137,140). Other key considerations to engage with patients and families in research include creating a welcome research environment, establishing expectations, providing support such as covering expenses, and acknowledging and promoting value of contributions (140). These principles and considerations can serve as a starting point to build partnerships with patient stakeholders.

Context of partnership with the Sibling Youth Advisory Council

In 2018, I established the Sibling Youth Advisory Council (SibYAC) as a research partner in my doctoral studies. In the literature, siblings with lived experiences of having a sibling with a disability have often described the importance of being involved in research (141). I recognized the value of POR, and as a trainee at the CanChild Centre for Childhood Disability Research, I saw how this centre embraced the culture of engagement with families in multiple projects (142). In addition, through my experiences as a graduate research assistant for the CHILD-BRIGHT READYorNot™ Brain-Based Disabilities Project, I was involved in facilitating and taking the lead on the co-creation of knowledge products with the Patient and Family Advisory Council. This council had valuable contributions that they brought to enhance the project. Based on these experiences, I wanted to ensure that I applied the values of POR to my dissertation. While there are knowledge gaps about understanding the experiences of siblings of individuals with a disability and synthesizing available resources in Canada support them, I wanted to ensure that this research was relevant and important to siblings. As I do not have the lived experiences of being a sibling of an individual with a disability, I recognize the value in incorporating the expertise of siblings of individuals with disabilities in research. My partnership with the SibYAC will help guide the research focus and enhance the quality of this dissertation.

Building my partnership with the Sibling Youth Advisory Council

Before starting my dissertation, I reached out to siblings of individuals with a disability to have a conversation, hear their stories, and share my ideas of my dissertation. Initially, I reached out to two siblings by word-of mouth who were already known to my graduate supervisory committee, for example, they were involved in other research, such as advisory councils, or with the CanChild Centre for Childhood Disability Research.

During the conversations with the two siblings, they were so passionate in sharing their stories and identified a need to further amplify the voices of siblings. For this reason, I then asked if they would be interested in being a part of research with my doctoral studies as a member of the Sibling Youth Advisory Council (SibYAC). Patient and family advisory councils is an approach that have been used to engage with stakeholders early in the research process (143). All siblings who I reached out expressed an interest in becoming a SibYAC member. I then invited them to attend a SibYAC meeting to learn more about the partnership and our work and they have continued to be engaged as a member of the SibYAC.

The SibYAC was initially comprised of two members. These two members were involved in the conception and design of one of the doctoral studies. Following my Comprehensive Examination during my second year of my doctoral studies in 2019, I wanted to expand the SibYAC to diversify perspectives and appropriate allocate time among members of the SibYAC. When engaging with patient and family partners, I had to consider the diversity and representation of those who engage, as well as appropriateness of time allocation (144). To incorporate these considerations from the literature, I then connected with three additional siblings through word-of-mouth; I reconnected with a sibling who I previously partnered in the Family Engagement in Research Certification of Completion Program (119), and my supervisor

(Dr. Jan Willem Gorter) connected me with two siblings who he knew through his research in which one sibling was a former postdoctoral fellow and another sibling was a member of the Patient and Family Advisory Council for CHILD-BRIGHT's READYorNot™ Brain-based Disabilities Project. At this point, I recognized that all members of the SibYAC identified as sisters, and I wanted to have diverse perspectives in my partnership with siblings. With the SibYAC, we discussed expanding the SibYAC to have additional siblings from different genders, including brothers, and one of the SibYAC partners introduced me to a brother. I followed a similar process to connect to these additional siblings as when I first started the SibYAC by having a conversation to learn about their experiences as a sibling and interest in joining the SibYAC. The SibYAC is currently comprised of six young adults (ages 21-28 years old) with 5 sisters and 1 brother. Five members are living in Ontario, Canada and one member is living in Alberta, Canada. All SibYAC members are siblings of a young person with a disability or chronic health condition including autism spectrum disorder, cerebral palsy, a genetic condition of CDKL5, and multiple sclerosis.

Continuing partnership with the Sibling Youth Advisory Council

My partnership with the SibYAC continued to be built as we collaborated on my doctoral studies. A key component that further enhanced our partnership was having clear and transparent communication. While there are multiple frameworks that can be used to guide patient and family engagement in research (139), the Patient Engagement Framework in the Strategy for Patient-Oriented Research from the Canadian Institutes of Health Research outlines common principles of inclusiveness, support, mutual respect, and co-build (17) which were applied throughout the partnership with the SibYAC. A qualitative study conducted by Black et al.

(2018) further identified the key considerations to be included when partnering with patients and families in research based on the perspectives of patients, families, and informal caregivers. The strategies that were used with the SibYAC based on these principles and considerations are presented in Table 3.

Table 3. Activities and tools that were conducted to build meaningful engagement with the SibYAC based on key principles identified from the Patient Engagement Framework (CIHR, 2019) and considerations from Black et al. (2018).

Consideration	**Description**	**Activities And Tools Conducted With The SibYAC**
Research Environment	To create a welcoming and warm environment, and provide a space for partners to share their perspectives.	• Terms of Reference (Supplementary File 2): This document outlines the background of my doctoral studies, membership of the SibYAC, roles that each SibYAC member might choose to have, how each SibYAC member would like to participate, and honoraria guidelines.
Expectations	To establish clear expectations about the roles and responsibilities, as well as provide closure about how feedback was incorporated into the project.	• Involvement Matrix (145): The Involvement Matrix was developed with experience experts (i.e., patients, their parents and/or family members), representatives of a patient organization BOSK (an association of persons with disabilities in the Netherlands), healthcare professionals, and researchers. This tool outlines the roles that individuals might want to have when engaging in different stages of preparation, execution, or implementation of the research project. These roles include being: a listener who is given information, a co-thinker who is asked to give opinions, an advisor who provides (un)solicited advice, a partner who works with researchers equally, or a decision-maker who takes initiatives and makes the final decisions. • Patient Engagement Tool (146): This tool outlines examples of ways that the SibYAC can be involved at different stages of research projects including plan the study, recruit and retain participants, do the study, analyze the results, and disseminate the results. This tool was helpful to be used with the Involvement Matrix as the SibYAC could identify ways that they might like to be involved in different phases of the research projects.
Support	To ensure that there were appropriate actions or attitudes that contribute to feelings of support with being a part of the research team.	• Group Meeting Rules and Facebook Group Rules (Supplementary File 3): Group rules were established to ensure that there was a welcoming and safe space for the SibYAC to share their perspectives. • Terms of Reference (Supplementary File 2): This document outlines information about honoraria.

Value	To provide opportunities for partners to apply their skill sets, incorporate the perspectives from partners based on their lived experiences, and acknowledge contributions from partners to the research projects.	• Individual check-in meetings: To be held with each SibYAC member once a year to discuss and reflect on the partnership. Each SibYAC member were asked to complete the 'Start, Stop, Continue' activity, which includes the questions of what activities should we start, stop, or continue doing. • Personal and group goals: Each SibYAC member shared their personal goals that motivated them to be a part of the SibYAC, as well as goals that they had for the collective SibYAC group. I wanted to ensure that not only were the valuable contributions of the SibYAC incorporated into my doctoral studies, but that there were also opportunities for the SibYAC to achieve their personal goals and for all of us to work as a team to achieve our collective SibYAC group goals.

Ongoing reflection and documentation is needed throughout the engagement with patient and family partners (147). As I continue to learn about how to engage and build my partnership with the SibYAC, I created opportunities to facilitate reflective practices with the SibYAC throughout the year during individual check-in meetings and team meetings. To provide further opportunities for reflection using an anonymous format, the Public and Patient Engagement Evaluation Tool (PPEET, 118) was administered to evaluate the self-reported perceptions from the SibYAC about their level of involvement in the doctoral research projects. The PPEET has undergone extensive usability testing and evaluation about its implementation among patient partners and professionals from healthcare organizations, and these studies were funded by the Ontario SPOR Support Unit (148,149). During the usability testing and evaluation of the PPEET, there were 131 participants with 11 participants who were 17-36 years old which is the same age range as SibYAC members (30).

Team meetings were held with the SibYAC once a month. Details about the structure of the team meetings are provided in Supplementary File 4. At the end of each meeting, a list of action items was provided if the SibYAC chose to be further involved with certain aspects of my doctoral studies, and these items could be completed by email or in a separate small group meeting. In addition to the involvement of the SibYAC in my doctoral studies, the SibYAC identified goals and initiatives that they would like to work on such as the co-creation of a website (150). The overall structure of the SibYAC continues to evolve based on feedback and insights shared by the SibYAC, as well as our individual and group goals. Funding support for my partnership with the SibYAC is provided in Supplementary File 5.

Overview and objectives of dissertation components

The overall aim of the program of research included in this dissertation was to deepen our understanding of the experiences of Canadian siblings of youth with disabilities during the transition to adulthood and to synthesize current resources to support them in their roles. This information can help to raise awareness about the unique and valuable roles that siblings have in the family. This dissertation is prepared as a sandwich thesis comprised of manuscripts that have been prepared, submitted, or published.

In **Chapter 2**, a review and qualitative document analysis was conducted to synthesize resources of siblings of youth with chronic health conditions to inform and support with healthcare management from organizations that are part of Children's Healthcare Canada. The objectives of this review are to identify and describe: 1) the types of resources currently available in Canada to provide both general information and specific healthcare management information about how siblings can provide support to their sibling with a chronic health condition; and 2) key topics discussed in resources created by siblings and families.

Chapter 3 describes the protocol of a scoping review about programs to prepare siblings for future roles to support their sibling with a neurodisability. The objective of the scoping review is to identify and map the characteristics and outcomes for participants in programs designed to prepare siblings in their future roles to support their brother or sister with a neurodisability. **Chapter 4** describes the results of the scoping review.

Chapter 5 describes the protocol of a qualitative case study titled, BrothErs and Sisters involvement in healthcare TranSition for youth wIth Brain-based disabilities (BEST SIBS) Study, with detailed descriptions of the methodology and techniques that were applied. The objectives of this study are to: 1) deepen our understanding of sibling roles, including any functions associated with healthcare transitions; and 2) identify the siblings' responsibilities in their relationship with their brother or sister with a neurodisability. The results of this qualitative case study are represented in **Chapter 6** and **Chapter 7**. In Chapter 6, the roles and responsibilities of siblings are described. In Chapter 7, findings represent the evolvement of sibling relationships through the developmental stages of childhood, adolescence, and emerging adulthood within a family environment.

Chapter 8 provides an overall discussion and conclusion of this dissertation. I share my personal reflections and learnings from my partnership with the SibYAC between 2018-2022. I also describe how this dissertation contributes to the development of knowledge about the relationships and support roles of siblings of youth with disabilities. Implications are shared with different stakeholders and future directions are suggested to enhance the level of support and types of resources for siblings of youth with disabilities for their current and future roles.

References

1. Stein REK, Jessop DJ. A noncategorical approach to chronic childhood illness. Public Health Reports. 1982;97(4):354–62.
2. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. Lancet Child Adolesc Health. 2018;2(3):223–8.
3. Zarrett N, Eccles J. The passage to adulthood: Challenges of late adolescence. New Directions for Youth Development. 2006;2006(111):13–28.
4. Stewart DA, Law MC, Rosenbaum P, Willms DG. A qualitative study of the transition to adulthood for youth with physical disabilities. Physical and Occupational Therapy in Pediatrics. 2001;21(4):3–21.
5. Blum RW. Introduction. Pediatrics. 2002;110(Supplement 3):1301.
6. Heath G, Farre A, Shaw K. Parenting a child with chronic illness as they transition into adulthood: A systematic review and thematic synthesis of parents' experiences. Patient Education and Counseling. 2017;100(1):76–92.
7. Nguyen T, Henderson D, Stewart D, Hlyva O, Punthakee Z, Gorter JW. You never transition alone! Exploring the experiences of youth with chronic health conditions, parents and healthcare providers on self-management. Child: Care, Health and Development. 2016;42(4):464–72.
8. Whiteman SD, McHale SM, Soli A. Theoretical perspectives on sibling relationships. Journal of Family Theory & Review. 2011;3(2):124–39.
9. Arnold CK, Heller T, Kramer J. Support needs of siblings of people with developmental disabilities. Intellectual and Developmental Disabilities. 2012;50(5):373–82.
10. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. Journal of Applied Research in Intellectual Disabilities. 2018;31(3):423–34.
11. Redquest BK, Tint A, Ries H, Goll E, Rossi B, Lunsky Y. Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2020;17(3):239–46.
12. Charlton JI. Nothing about us without us: Disability oppression and empowerment. Berkeley: University of California Press; 1998.
13. Paul T. "Nothing about us without us": Toward patient- and family-centered care. AMA Journal of Ethics. 2016;18(1):3–5.
14. Stack E, McDonald KE. Nothing about us without us: Does action research in developmental disabilities research measure up? Journal of Policy and Practice in Intellectual Disabilities. 2014;11(2):83–91.
15. Stack EE, McDonald K. We are "both in charge, the academics and self-advocates": Empowerment in community-based participatory research. Journal of Policy and Practice in Intellectual Disabilities. 2018;15(1):80–9.
16. Canadian Institutes of Health Research. Strategy for Patient Oriented Research. Patient engagement framework [Internet]. 2014. Available from: http://www.cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf
17. Government of Canada - Canadian Institutes of Health Research. Strategy for Patient-Oriented Research [Internet]. 2021 [cited 2021 Nov 7]. Available from: https://cihr-irsc.gc.ca/e/41204.html
18. Perrin JM, Bloom SR, Gortmaker SL. The increase of childhood chronic conditions in the United States. JAMA. 2007;297(24):2755.

19. Newacheck PW, Strickland B, Shonkoff JP, Perrin JM, McPherson M, McManus M, et al. An epidemiologic profile of children with special health care needs. Pediatrics. 1998;102(1):117–23.
20. Knecht C, Hellmers C, Metzing S. The perspective of siblings of children with chronic illness: a literature review. Journal of Pediatric Nursing. 2015;30(1):102–16.
21. Kids Brain Health Network. Neurodisabilities: Canadian Prevalence [Internet]. 2021 [cited 2022 Jan 18]. Available from: https://kidsbrainhealth.ca/index.php/portfolio-items/neurodisabilities-canadian-prevalence/
22. Morris C, Janssens A, Tomlinson R, Williams J, Logan S. Towards a definition of neurodisability: a Delphi survey. Developmental Medicine & Child Neurology. 2013;55(12):1103–8.
23. Miller AR, Rosenbaum P. Perspectives on "disease" and "disability" in child health: The case of childhood neurodisability. Frontiers in Public Health. 2016;4:1–13.
24. King S, Teplicky R, King G, Rosenbaum P. Family-centered service for children with cerebral palsy and their families: A review of the literature. Seminars in Pediatric Neurology. 2004;11(1):78–86.
25. Gibson BE, Teachman G, Hamdani Y. Rethinking "Normal Development" in Children's Rehabilitation. In: Rethinking Rehabilitation. CRC Press; 2015. p. 90–103.
26. Hammell KW, Jarus T, Bulk LY, Collins B, Grenier ML, Mahipaul S. Letter to the editor. Vol. 88, Canadian Journal of Occupational Therapy. SAGE Publications Inc.; 2021. p. 279–82.
27. World Health Organization. International Classification of Functioning, Disability and Health (ICF). Geneva, Switzerland: World Health Organization; 2001.
28. Statistics Canada. Census in Brief: Portrait of children's family life in Canada in 2016. 2017.
29. Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. American Psychologist. 2000;55(5):469–80.
30. Gorter JW, Stewart D, Woodbury-Smith M. Youth in transition: care, health and development. Child: Care, Health and Development. 2011;37(6):757–63.
31. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Canadian Journal of Community Mental Health. 2014;33(1):45–61.
32. Scal P, Evans T, Blozis S, Okinow N, Blum R. Trends in transition from pediatric to adult health care services for young adults with chronic conditions. Journal of Adolescent Health. 1999;24(4):259–64.
33. Freeman M, Stewart D, Cunningham CE, Gorter JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. Journal of Transition Medicine. 2018;1(1).
34. Palisano RJ, Di Rezze B, Stewart D, Rosenbaum PL, Hlyva O, Freeman M, et al. Life course health development of individuals with neurodevelopmental conditions. Developmental Medicine & Child Neurology. 2017;59(5):470–6.
35. Canadian Paediatric Society. Positon Statement (AH 2007-01). Transition to adult care for youth with special health care needs. Paediatric Child Health. 2007;12(9):785–8.
36. Young NL. The transition to adulthood for children with cerebral palsy. Journal of Pediatric Orthopaedics. 2007;27(4):476–9.

37. Young NL, Gilbert TK, McCormick A, Ayling-Campos A, Boydell K, Law M, et al. Youth and young adults with cerebral palsy: Their use of physician and hospital services. Archives of Physical Medicine and Rehabilitation. 2007;88(6):696–702.
38. Young NL, McCormick A, Mills W, Barden W, Boydell K, Law M, et al. The transition study: a look at youth and adults with cerebral palsy, spina bifida, and acquired brain injury. Phys Occup Ther Pediatr. 2006;26(4):25–45.
39. Murphy NA, Hoff C, Jorgensen T, Norlin C, Young PC. Costs and complications of hospitalizations for children with cerebral palsy. Pediatric Rehabilitation. 2006 Jan 10;9(1):47–52.
40. Stewart D. Transition to adult services for young people with disabilities: Current evidence to guide future research. Developmental Medicine and Child Neurology. 2009;51(Supp. 4):169–73.
41. Nguyen T, Gorter JW. Use of the international classification of functioning, disability and health as a framework for transition from paediatric to adult healthcare. Child: Care, Health and Development. 2014;40(6):759–61.
42. Nguyen T, Henderson D, Stewart D, Hlyva O, Punthakee Z, Gorter JW. You never transition alone! Exploring the experiences of youth with chronic health conditions, parents and healthcare providers on self-management. Child: Care, Health and Development. 2016;42(4):464–72.
43. American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians-American Society of Internal Medicine. A Consensus Statement on Health Care Transitions for Young Adults. Pediatrics. 2002;110(Supplement 3):1304–6.
44. Toulany A, Gorter JW, Harrison ME, Canadian Pediatric Society, Adolescent Health Committee. A call for action: Recommendations to improve transition to adult care for youth with complex health care needs [Internet]. 2022 [cited 2022 Apr 15]. Available from: https://cps.ca/en/documents/position/transition-to-adult-care-for-youth
45. Lunsky Y, Robinson S, Blinkhorn A, Ouellette-Kuntz H. Parents of adults with intellectual and developmental disabilities (IDD) and compound caregiving responsibilities. Journal of Child and Family Studies. 2017;26(5):1374–9.
46. Haley WE, Perkins EA. Current status and future directions in family caregiving and aging people with intellectual disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2004;1(1):24–30.
47. Kim HW, Greenberg JS, Seltzer MM, Krauss MW. The role of coping in maintaining the psychological well-being of mothers of adults with intellectual disability and mental illness. Journal of Intellectual Disability Research. 2003;47(4–5):313–27.
48. Novak-Pavlic M, Abdel Malek S, Rosenbaum P, Macedo LG, Di Rezze B. A scoping review of the literature on grandparents of children with disabilities. Disability and Rehabilitation. 2021;1–15.
49. Rosen D. Between two worlds: Bridging the cultures of child health and adult medicine. Journal of Adolescent Health. 1995;17(1):10–6.
50. American Academy of Pediatrics Committee. Family-centered care and the pediatrician's role. Pediatrics. 2003;112(3 I):691–6.
51. Bamm EL, Rosenbaum P. Family-centered theory: origins, development, barriers, and supports to implementation in rehabilitation medicine. Arch Phys Med Rehabil. 2008;89(8):1618–24.
52. Briar-Lawson K, Lawson HA, Hennon CB. Family-centered policies and practices:

International implications. New York: Columbia University Press; 2001.
53. Rosenbaum P, King S, Law M, King G, Evans J. Family-centred service. Physical & Occupational Therapy In Pediatrics. 1998;18(1):1–20.
54. Gall C, Kingsnorth S, Healy H, Gall C, Healy H. Growing up ready: A shared management approach. Physical & Occupational Therapy in Pediatrics. 2009;26(4):47–62.
55. Reiss JG, Gibson RW, Walker LR. Health care transition: Youth, family, and provider perspectives. Pediatrics. 2005;115(1):112–20.
56. Rehm RS, Fuentes-Afflick E, Fisher LT, Chesla CA. Parent and youth priorities during the transition to adulthood for youth with special health care needs and developmental disability. Advances in Nursing Science. 2012;35(3).
57. Lozano P, Houtrow A. Supporting self-management in children and adolescents with complex chronic conditions. Pediatrics. 2018;141(Supplement 3):S233–41.
58. Sanner C, Russell LT, Coleman M, Ganong L. Half-sibling and stepsibling relationships: A systematic integrative review. Journal of Family Theory & Review. 2018 Dec 1;10(4):765–84.
59. Statistics Canada. Census Profile, 2016 Census [Internet]. [cited 2022 Apr 15]. Available from: https://www12.statcan.gc.ca/census-recensement/2016/dp-pd/prof/details/page.cfm?Lang=E&Geo1=PR&Code1=01&Geo2=PR&Code2=01&SearchText=Canada&SearchType=Begins&SearchPR=01&B1=Families, households and marital status&TABID=1&type=0
60. Dunn S, Zwicker J. It's time for Canada to measure up on kids with disabilities. The Star. 2017 Nov 12;
61. Government of Canada. Disability in Canada: A 2006 Profile [Internet]. [cited 2022 Apr 15]. Available from: https://www.canada.ca/en/employment-social-development/programs/disability/arc/disability-2006.html
62. Shikako K, Gonzalez M, Lai J, Movahed M, Severino SM, Martens R, et al. Children with disabilities in Canada: Alternative report for the 5th/6th review of Canada under the Convention on the Rights of the Child [Internet]. 2020. Available from: https://www.childhooddisability.ca/wp-content/uploads/2020/03/AlternativeREPORT.pdf
63. Hodapp RM, Sanderson KA, Meskis SA, Casale EG. Adult siblings of persons with intellectual disabilities: Past, present, and future. International Review of Research in Developmental Disabilities. 2017;53:163–202.
64. Bank P, Kahn M. The sibling bond. New York: Basic Books; 1982.
65. Atkin K, Tozer R. Personalisation, family relationships and autism: Conceptualising the role of adult siblings. Journal of Social Work. 2014;14(3):225–42.
66. Corsano P, Musetti A, Guidotti L, Capelli F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. Journal of Intellectual and Developmental Disability. 2017;42(2):151–61.
67. Latta A, Rampton T, Rosemann J, Peterson M, Mandleco B, Dyches T, et al. Snapshots reflecting the lives of siblings of children with autism spectrum disorders. Child: Care, Health and Development. 2014;40(4):515–24.
68. Scharf M, Shulman S, Avigad-Spitz L. Sibling relationships in emerging adulthood and in adolescence. Journal of Adolescent Research. 2005;20(1):64–90.
69. Neyer FJ. Twin relationships in old age: A developmental perspective. Journal of Social and Personal Relationships. 2002;19:55–177.
70. McHale SM, Updegraff KA, Whiteman SD. Sibling relationships and influences in

childhood and adolescence. Journal of Marriage and Family. 2012;74(5):913–30.

71. Stoneman Z, Brody GH. Sibling temperaments, conflict, warmth, and role asymmetry. Child Development. 1993;64(6):1786–800.

72. Windle M, Lerner RM. Reassessing the dimensions of temperamental individuality across the life span: The revised Dimensions of Temperament Survey (DOTS-R). Journal of Adolescent Research. 1986;1(2):213–29.

73. Thomas A, Chess S. Genesis and evolution of behavioral disorders: From infancy to early adult life. Annual Progress in Child Psychiatry & Child Development. 1985;140–58.

74. Stoneman Z, Brody GH, Churchill SL, Winn LL. Effects of residential instability on Head Start children and their relationships with older siblings: Influences of child emotionality and conflict between family caregivers. Child Development. 1999;70(5):1246–62.

75. Tschann JM, Kaiser P, Chesney MA, Alkon A, Boyce WT. Resilience and vulnerability among preschool children: Family functioning, temperament, and behavior problems. J Am Acad Child Adolesc Psychiatry. 1996;35(2):184–92.

76. Wertlieb D, Weigel C, Springer T, Feldstein M. Temperament as a moderator of children’s stressful experiences. American Journal of Orthopsychiatry. 1987 Apr;57(2):234–45.

77. Stoneman Z. Supporting positive sibling relationships during childhood. Mental Retardation and Developmental Disabilities Research Reviews. 2001;7(2):134–42.

78. Hodapp RM, Urbano RC. Adult siblings of individuals with Down syndrome versus with autism: Findings from a large-scale US survey. Journal of Intellectual Disability Research. 2007;51(12):1018–29.

79. Orsmond GI, Seltzer MM. Siblings of individuals with autism or Down syndrome: Effects on adult lives. Journal of Intellectual Disability Research. 2007;51(9):682–96.

80. Doody MA, Hastings RP, O’Neill S, Grey IM. Sibling relationships in adults who have siblings with or without intellectual disabilities. Research in Developmental Disabilities. 2010;31(1):224–31.

81. Rossetti Z, Hall S. Adult sibling relationships with brothers and sisters with severe disabilities. Research and Practice for Persons with Severe Disabilities. 2015;40(2):120–37.

82. Giallo R, Gavidia-Payne S, Minett B, Kapoor A. Sibling voices: The self-reported mental health of siblings of children with a disability. Clinical Psychologist. 2012;16(1):36–43.

83. Petalas MA, Hastings RP, Nash S, Dowey A, Reilly D. “I like that he always shows who he is”: The perceptions and experiences of siblings with a brother with autism spectrum disorder. International Journal of Disability, Development and Education. 2009;56(4):381–99.

84. Moyson T, Roeyers H. The quality of life of siblings of children with autism spectrum disorder. Exceptional Children. 2011;78(1):41–55.

85. Meltzer A. ‘I couldn’t just entirely be her sister’: the relational and social policy implications of care between young adult siblings with and without disabilities. Journal of Youth Studies. 2017;20(8):1013–27.

86. Mauldin L, Saxena M. Mutual exchange: Caregiving and life enhancement in siblings of individuals with developmental disabilities. Journal of Family Issues. 2018;39(8):2232–64.

87. Dunn J, Slomkowski C, Bcardsall L, Rende R. Adjustment in middle childhood and early adolescence: Links with earlier and contemporary sibling relationships. Journal of Child

Psychology and Psychiatry. 1994;35(3):491–504.
88. Akiyama H, Elliott K, Antonucci TC. Same-sex and cross-sex relationships. The Journals of Gerontology Series B: Psychological Sciences and Social Sciences. 1996;51B(6):P374–82.
89. Burbidge J, Minnes P. Relationship quality in adult siblings with and without developmental disabilities. Family Relations. 2014;63(1):148–62.
90. Hodapp RM, Urbano RC, Burke MM. Adult female and male siblings of persons with disabilities: Findings from a national survey. Intellectual and Developmental Disabilities. 2010;48(1):52–62.
91. Orsmond GI, Seltzer MM. Brothers and sisters of adults with mental retardation: Gendered nature of the sibling relationship. American Journal on Mental Retardation. 2000;105(6):486–508.
92. Cuskelly M. Contributors to adult sibling relationships and intention to care of siblings of individuals with Down syndrome. American Journal on Intellectual and Developmental Disabilities. 2016;121(3):204–18.
93. Furman W, Buhrmester D. Children's perceptions of the qualities of sibling relationships. Child Development. 1985;56(2):448.
94. Buhrmester D, Furman W. Perceptions of sibling relationships during middle childhood and adolescence. Child Development. 1990;61(5):1387–98.
95. Tucker CJ, McHale SM, Crouter AC. Conditions of sibling support in adolescence. Journal of Family Psychology. 2001;15(2):254–71.
96. Mandleco B, Webb AEM. Sibling perceptions of living with a young person with Down syndrome or autism spectrum disorder: An integrated review. Journal for Specialists in Pediatric Nursing. 2015;20(3):138–56.
97. Meyer D, Holl E. The sibling survival guide: Indispensable information for brothers and sisters of adults with disabilities. Bethesda, MD: Woodbine House; 2014.
98. Rivers JW, Stoneman Z. Sibling relationships when a child has autism: marital stress and support coping. Journal of Autism and Developmental Disorders. 2003;33(4):383–94.
99. Shivers CM, Dykens EM. Adolescent siblings of individuals with and without intellectual and developmental disabilities: Self-reported empathy and feelings about their brothers and sisters. American Journal on Intellectual and Developmental Disabilities. 2017;122(1):62–77.
100. Chambers CR, Hughes C, Carter EW. Parent and sibling perspectives on the transition to adulthood. Education and Training in Developmental Disabilities. 2004;39(2):79–94.
101. Burke MM, Patton KA, Taylor JL. Family support: A review of the literature on families of adolescents with disabilities. Journal of Family Social Work. 2016;19(4):252–85.
102. Cutrona CE. Social support principles for strengthening families: messages from the USA. In: Canavan J, Dolan P, Pinkerton J, editors. Family Support: Direction from Diversity. London: Jessica Kingsley; 2000. p. 103–22.
103. Schaefer C, Coyne JC, Lazarus RS. The health-related functions of social support. Journal of Behavioral Medicine. 1981;4(4):381–406.
104. Bigby C. Parental Substitutes. Journal of Gerontological Social Work. 1998;29(1):3–21.
105. Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. Intellectual and Developmental Disabilities. 2009;47(3):208–19.
106. Holl E, Morano CL. Supporting the next generation of caregivers: Service use and needs

of adult siblings of individuals with intellectual disability. Inclusion. 2014;2(1):2–16.

107. Davys D, Mitchell D, Haigh C. Futures planning, parental expectations and sibling concern for people who have a learning disability. Journal of Intellectual Disabilities. 2010;14(3):167–83.

108. D'Arcy F, Flynn J, McCarthy Y, O'Connor C, Tierney E. Sibshops. An evaluation of an interagency model. Journal of Intellectual Disabilities. 2005;9(1):43–57.

109. Opening Hearts. SibTeens [Internet]. 2019 [cited 2021 Jun 2]. Available from: https://www.openinghearts.ca/sibteens/

110. Knox M, Bigby C. Moving towards midlife care as negotiated family business: Accounts of people with intellectual disabilities and their families "Just getting along with their lives together." International Journal of Disability, Development and Education. 2007;54(3):287–304.

111. The Sibling Collaborative. Understanding the sibling experience: sibling needs assessment survey 2017 [Internet]. 2018 [cited 2021 Jun 2]. p. 1–19. Available from: http://cdn.agilitycms.com/partners-for-planning/Understanding the Sibling Experience - March 2018 - Final.pdf

112. Rawson H. "I'm going to be here long after you've gone" - sibling perspectives of the future. British Journal of Learning Disabilities. 2010;38(3):225–31.

113. Creswell JW, Poth CN. Philosophical assumptions and interpretive frameworks. In: Qualitative Inquiry & Research Design: Choosing Among Five Approaches. 4th ed. Thousand Oaks, CA, US: Sage Publications Inc.; 2018. p. 15–40.

114. Denzin NK, Lincoln YS. Introduction: The discipline and practice of qualitative research. In: The SAGE handbook of qualitative research. 4th ed. Thousand Oaks, CA, US: Sage Publications Inc.; 2011. p. 1–19.

115. Mertens DM. Research and evaluation in education and psychology: Integrating diversity with quantitative, qualitative, and mixed methods. 4th ed. Thousand Oaks, California: Sage Publications Inc.; 2015.

116. Han H. The influence of differences in values between Chinese and Weestern cultures on communication modes. Advances in Social Science, Education and Humanities Research. 2018;185.

117. Hospice Toronto. Young carers program [Internet]. [cited 2022 May 8]. Available from: https://www.ycptoronto.com/

118. CanChild Centre for Childhood Disability Research. MyTransition App [Internet]. [cited 2022 Apr 25]. Available from: https://www.canchild.ca/en/research-in-practice/current-studies/apply-the-mytransition-app-in-transition-applyit-study/mytransition-app

119. CanChild Centre for Childhood Disability Research. Family Engagement in Research Course [Internet]. 2022 [cited 2022 Feb 8]. Available from: https://www.canchild.ca/en/research-in-practice/family-engagement-in-research-course

120. Gorter JW, Amaria K, Kovacs A, Rozenblum R, Thabane L, Galuppi B, et al. CHILD-BRIGHT READYorNot Brain-Based Disabilities Trial: Protocol of a randomised controlled trial (RCT) investigating the effectiveness of a patient-facing e-health intervention designed to enhance healthcare transition readiness in youth. BMJ Open. 2021;11:48756.

121. Connelly LM. Use of theoretical frameworks in research. MedSurg Nursing. 2014;23(3):187–9.

122. Meleis AI, Sawyer LM, Im EO, Messias DK, Schumacher K. Experiencing transitions: an

emerging middle-range theory. Advances in Nursing Science. 2000;23(1):12–28.

123. Dew A, Boydell KM. Knowledge translation: bridging the disability research-to-practice gap. Research and Practice in Intellectual and Developmental Disabilities. 2017;4(2):142–57.

124. Powers JL, Tiffany JS. Engaging youth in participatory research and evaluation. Journal of Public Health Management and Practice. 2006;12(SUPPL. 6):79–87.

125. Brett J, Staniszewska S, Mockford C, Herron-Marx S, Hughes J, Tysall C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. Health Expectations. 2014;17(5):637–50.

126. Domecq JP, Prutsky G, Elraiyah T, Wang Z, Nabhan M, Shippee N, et al. Patient engagement in research: A systematic review. BMC Health Services Research. 2014;14(1):89.

127. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: a scoping review of the 'how' and 'what' of patient engagement in health research. Health Research Policy and Systems. 2018;16(1):5.

128. Bailey S, Boddy K, Briscoe S, Morris C. Involving disabled children and young people as partners in research: A systematic review. Child: Care, Health and Development. 2015;41(4):505–14.

129. Bruce SM, Parker AT. Young deafblind adults in action: Becoming self-determined change agents through advocacy. American Annals of the Deaf. 2012;157(1):16–26.

130. Dedding C. Delen in macht en onmacht. Universiteit van Amsterdam; 2009.

131. Lightfoot J, Sloper P. Having a say in health: involving young people with a chronic illness or physical disability in local health services development. Children & Society. 2003;17(4):277–90.

132. Canter KS, Roberts MC. A systematic and quantitative review of interventions to facilitate school reentry for children with chronic health conditions. Journal of Pediatric Psychology. 2012;37(10):1065–75.

133. Dudley L, Gamble C, Preston J, Buck D, Hanley B, Williamson P, et al. What difference does patient and public involvement make and what are its pathways to impact? Qualitative study of patients and researchers from a cohort of randomised clinical trials. Jepson R, editor. PLOS ONE. 2015;10(6):e0128817.

134. Vale CL, Thompson LC, Murphy C, Forcat S, Hanley B. Involvement of consumers in studies run by the Medical Research Council Clinical Trials Unit: Results of a survey. Trials. 2012;13(1):9.

135. Curtin C. Eliciting children's voices in qualitative research. American Journal of Occupational Therapy. 2001;55(3):295–302.

136. Barker J, Weller S. "Is it fun?" Developing children centred research methods. International Journal of Sociology and Social Policy. 2003;23(1):23–58.

137. van Schelven F, Boeije H, Mariën V, Rademakers J. Patient and public involvement of young people with a chronic condition in projects in health and social care: A scoping review. Health Expectations. 2020;23(4):789–801.

138. Arnstein SR. A Ladder Of Citizen Participation. J Am Inst Plann. 1969;35(4):216–24.

139. Nguyen T, Palisano RJ, Graham I. Perspectives and experiences with engaging youth and families in research. Physical and Occupational Therapy in Pediatrics. 2019;39(3):310–23.

140. Black A, Strain K, Wallsworth C, Charlton SG, Chang W, McNamee K, et al. What constitutes meaningful engagement for patients and families as partners on research

teams? Journal of Health Services Research and Policy. 2018;23(3):158–67.

141. Richardson SLL, Jordan LS. Qualitative inquiry of sibling relationships: reinforcement of disability devaluation through the exclusion of voices. Disability and Society. 2017;32(10):1534–54.

142. Pozniak K, Buchanan F, Cross A, Crowson J, Galuppi B, Grahovac D, et al. Building a culture of engagement at a research centre for childhood disability. Research Involvement and Engagement. 2021;7(1):78.

143. Harrison JD, Auerbach AD, Anderson W, Fagan M, Carnie M, Hanson C, et al. Patient stakeholder engagement in research: A narrative review to describe foundational principles and best practice activities. Health Expectations. 2019;22(3):307–16.

144. Esmail L, Moore E, Rein A. Evaluating patient and stakeholder engagement in research: moving from theory to practice. Journal of Comparative Effectiveness Research. 2015;4(2):133–45.

145. Smits DW, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. Research Involvement and Engagement. 2020;6(1):30.

146. Ontario Brain Institute. Ways community members can participate in the stages of research. 2019.

147. Staley K. ‘Is it worth doing?’ Measuring the impact of patient and public involvement in research. Research Involvement and Engagement. 2015;1(1):1–10.

148. Abelson J, Li K, Wilson G, Shields K, Schneider C, Boesveld S. Supporting quality public and patient engagement in health system organizations: development and usability testing of the Public and Patient Engagement Evaluation Tool. Health Expectations. 2016;19(4):817–27.

149. Abelson J, Tripp L, Kandasamy S, Burrows K. Supporting the evaluation of public and patient engagement in health system organizations: Results from an implementation research study. Health Expectations. 2019;22(5):1132–43.

150. CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study [Internet]. 2021 [cited 2022 Feb 4]. Available from: https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study

Supplementary File 1. Learnings to build knowledge and experience in partnering with patient and family partners in research.

Learnings from the Family Engagement in Research Certificate of Completion Program

This program was hosted by McMaster University Centre for Continuing Education, CanChild Centre for Childhood Disability Research, and Kids Brain Health Network (116). This course is available for researchers such as graduate students, research coordinators, investigators, and clinician-researchers, as well as families such as parents, siblings and grandparents, who have an interest in child neurodevelopmental research. Through this course, I had the opportunity to learn about the foundational principles of family engagement including its importance, barriers and facilitators to engagement, ethics of engagement, as well as tools and resources to support and evaluate engagement activities. One of the key learning experiences was the experiential component, in which I partnered with a sibling with lived experiences of having a brother with a genetic condition and a PhD student in biomedical sciences. We partnered together to co-develop an infographic of strategies to engage in biomedical and clinical research projects. It was through this experiential component that I learned how there are multiple ways to engage with partners in research.

Learnings from the partnership with the Patient and Family Advisory Council and READYorNot™ Brain-Based Disabilities Project

During my PhD studies, I also had the opportunity to work as a graduate research assistant (Sept 2017 – Dec 2021) with my supervisor, Dr. Gorter, as a Co-Principal Investigator for the CHILD-BRIGHT: Child Health Initiatives Limiting Disability – Brain Research Improving Growth and Health Trajectories, with the project titled, READiness in Youth fOR traNsition Out of pediaTric Care ('READYorNot' Brain-Based Disabilities) funded by the Canadian Institutes of Health Research – Strategy for Patient-Oriented Research. This project aims to develop an eHealth intervention, the 'MyREADY Transition™ BBD' App, and evaluate whether this App supports youth to enhance transition readiness from pediatric to adult healthcare (117). In my role as a graduate research assistant, I provided support with facilitating meetings with the Patient and Family Advisory Council, including the co-development of research products, such as a series of recruitment videos to explain research in lay language to youth (148), as well as co-presentations at conferences. In September 2019, I became a member of the CHILD-BRIGHT Training Committee to plan different initiatives, to help others learn about patient-oriented research. Based on my experiences about patient-oriented research, I wanted to apply these principles to my doctoral studies.

Supplementary File 2. Terms of Reference.

Sibling Youth Advisory Council (SibYAC)
Terms of Reference

1. Background

Youth with a disability may experience challenges when they transition from pediatric to adult healthcare. Youth can look to their families for support during healthcare transition. Siblings are also a part of the family, who often understand the unique needs and concerns of their brother or sister with a disability. Siblings can play different roles, such as a friend, role model, or mentor. There should be increased awareness about the potential roles that siblings can play during healthcare transition. Currently, there are limited resources for siblings who are 14 years or older to support their brother or sister prepare for healthcare transition.

The overall aim of this doctorate research program is to understand the experiences of siblings who have a brother or sister with a disability and is in the process of preparing for the transition to adulthood.

2. Membership

The SibYAC will consist of young adult siblings who have a brother or sister with a disability, and we will seek to have representation of siblings with diverse backgrounds (e.g., age, gender) across Canada.

3. Role

Sibling Partners may choose to take on any of the following roles and responsibilities for different tasks of each study[1]:

- ***Listeners*** are given information about the project from the research team.
- ***Co-thinkers*** are asked to give their opinion.
- ***Advisors*** gives (un)solicited advice.
- ***Partners*** works as an equal partner with other members of the research team.
- ***Decision-makers*** take initiatives themselves.

---

[1] Smits, D. W., van Meeteren, K., Klem, M., Alsem, M., & Ketelaar, M. (2020). Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. *Research involvement and engagement*, *6*(1), 1-7.

4. Participation

SibYAC members are invited to attend group monthly meetings and communicate regularly in various formats (e.g., emails, individual check-in meetings, Facebook group). To become a member of the SibYAC, you must have: a) access to a telephone and/or computer connected to the Internet; and b) be able to communicate in English. A toll free (no cost) number will be provided for you to connect and meet with us by telephone.

You may decide to leave the SibYAC at any time. You may also choose to take a temporary leave from the SibYAC, please communicate this to Linda Nguyen, NGUYEL7@mcmaster.ca. Below is a description of each study in the doctorate research program and examples of how SibYAC members can contribute to each study.

Study 1: Knowledge Inquiry (In Progress)

A scoping review will be conducted to summarize information from the literature about programs to support siblings. Since 2018, existing SibYAC members have contributed to the development of the research question, study design, and reading key articles from the literature. Moving forward, SibYAC members will have the opportunity to extract data information from selected articles, analyze extracted data, and discuss relevance and implication of results based on experiences.

Study 2: Knowledge Inquiry (Completed)

A qualitative document analysis will be conducted to identify resources that are available for siblings of individuals with a chronic health condition across children's hospitals and treatment centres across Canada. Since 2018, existing SibYAC members have contributed to the development of the research question. Similar to Study 1, the SibYAC will have the opportunity to extract data information from selected articles, analyze extracted data, and discuss relevance and implications of results based on experiences.

Study 3: Knowledge Contextualization (In Progress)

A qualitative case study, titled BrothErs and Sisters involvement in healthcare TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study, to understand siblings' roles and responsibilities to their brother or sister with a childhood-onset neurodisability during healthcare transition. Since 2018, existing SibYAC members have contributed to the development of the research question, study design, recruitment materials, as well as pilot testing the interview guide. Moving forward, SibYAC members can choose to contribute to the study by sharing recruitment materials to recruit participants, analyze data from study interviews, and discuss the relevance and implications of the study results.

Study 4: Knowledge Product/Tool (To Be Conducted)

Based on findings from Study 1 and Study 2, a Sibling Kit will be co-created with the SibYAC as a resource to support siblings during the healthcare transition of their brother or sister with a disability. The SibYAC will be involved throughout all stages of the study, including the preparation, execution, and implementation stages.

Knowledge Translation and Implementation (Ongoing)

All studies will include a knowledge translation and implementation phase, where SibYAC members can share the research findings with the community. For example, there will be opportunities for SibYAC members to co-present at conferences, co-write lay language summaries about the study findings, and co-write publications.

5. Honoraria

The role of a SibYAC member is voluntary, but we wish to provide an honorarium as an appreciation of your valuable contributions of your time, effort, commitment, and expertise. You may decline to participate in any SibYAC activities, however, you must attend at least 3 meetings within a 6-month period in order to receive the full compensation. Requests can be made for partial, full, or additional payment that aligns with the contributions from each SibYAC member in a 6-month period.

There are different forms of honoraria to recognize the roles and contributions of each SibYAC member.

Type of Honoraria	**Commitment**	**Responsibility**	**Roles**	**Suggested Honoraria Amount***
Annual honoraria for commitments to the doctorate research program	Availability by email; willing and able to participate in some meetings by phone	Shares advice and opinions for the study, may choose to work as an equal partner or decision-maker	Listener Co-thinker Advisor Partner Decision-maker	$250.00 per year (for all roles)
Additional honoraria for extra activities on behalf of the doctorate research program	Examples of additional activities may include: • co-presenting a webinar, conference, or podcast; • co-writing an article (e.g., newsletter, publication); • writing a personal blog or story	May include: • preparing materials (e.g., slides and script), recording or delivering the presentation; • review and provide feedback on written materials (e.g., drafted abstract) • writing sections of an article (e.g., newsletter, a personal story or blog, publication)	Listener Co-thinker Advisor	Information can be shared but an additional honorarium would not be provided.
			Partner Decision-maker	$25-75 per activity • Personal blog, story or newsletter - $25 • Conference presentations - $50 • Webinar, podcast or co-publication - $75 *Note: The honorarium amount can be used towards the cost of a group activity instead (e.g., virtual social gathering).
Honoraria for research training activities	Completion of training activities (e.g., Strategy for Patient-Oriented Research training such as the "Patient-Oriented Research Curriculum in Child	Completion of training activities.	Listener	$75.00

	Health" or PORCCH)			
Honoraria for personal development activities	Participation in personal development activities (e.g., participation in a course to learn new skills, such as oral communication or data analysis; attending webinars or workshops to learn about developing a research career).	Personal development activities can be further discussed.	Partner Decision-maker	$75.00

*Note: A suggested honorarium amount is provided, but the honorarium for each activity can be discussed with Linda Nguyen.

6. Applying to the SibYAC

We hope that your role as a member of the SibYAC will be a rewarding and valuable experience for you.

However, we understand if you choose not to be a part of the SibYAC. If you are **NOT interested** in joining the SibYAC but would like to be invited to participate in a study, please us know by email (NGUYEL7@mcmaster.ca) and we will contact you later.

If you are interested in becoming a member of the SibYAC, please complete the following form and save the completed form for your records. Please send a copy of your completed form attached to an email to Linda Nguyen, NGUYEL7@mcmaster.ca.

Name (first and last):	Click here to enter text.		
Email Address:	Click here to enter text.		
Mailing Address:	Click here to enter text.		
Telephone Number *(with area code):*	Click here to enter text.		
Which best describes where you live?	**Province:**	Click here to enter text.	
	Rural/Town ☐	**City** ☐	**Northern/Remote** ☐
Which ethic/cultural background do you identify with?	Click here to enter text.		
Your age:	Click here to enter text.		
Age of your sibling:	Click here to enter text.		
Do you have a neurodevelopmental disability (either diagnosed or not diagnosed)?	**YES** ☐	**NO** ☐	
	If yes, please specify: Click here to enter text.		
Do you have a sibling with a neurodevelopmental disability (either diagnosed or not diagnosed?	**YES** ☐	**NO** ☐	
	If yes, please specify: Click here to enter text.		
What is your or your sibling's functional limitations?	**mobility** ☐		
	vision ☐		
	communication ☐		
	learning ☐		
	attention ☐		
	other: Click here to enter text.		
Please tell us a little about why you'd like to join the SibYAC (optional).	Click here to enter text.		

*Adapted from the Terms of Reference for the Patient and Family Advisory Council (PFAC) for the CHILD-BRIGHT READYorNot™ Brain-Based Disabilities Project.

Supplementary File 3. Group Meeting Rules and Facebook Group Rules.

Sibling Youth Advisory Council (SibYAC)
Group Meeting Rules

1. **Treat people the same as you would face-to-face**
   Remember that there is a person behind the computer screen, and we should remember to treat others with respect in ways that we would in person.

2. **Please remember not to share sensitive information**
   The facilitator (Linda Nguyen) is not responsible for the outcomes of information that is shared in this group, so please use your discretion for the information that you share during meetings. If you are unsure of information to post, you are welcome to check in with the facilitator.

3. **Engagement with others**
   This is a space for us to build a community, share information, and connect socially. Please consider the intent of your posts and chat with others. We are your community, so please feel free to contribute during meetings.

4. **No medical advice please**
   We encourage open communication and sharing of information. Please note that due to legal concerns, there will be no medical advice given.

5. **Your privacy matters**
   Please remember that this is a safe space for everyone to share their experiences and stories. Nothing that you share (photos, personal information, comments, anecdotes) may be repeated or used by others in another setting or publicly without your expressed consent. If you are asked for permission, you are not obligated to agree to it. It is entirely up to you what you decide to share.

6. **Research information**
   For information related to Linda Nguyen's doctoral studies, please remember that the research is being conducted and refrain from sharing the research with others before having a discussion in this group.

7. **Data collection may happen on occasion**
   Information in this group may be used for data collection and/or publication purposes. This is never done without your permission. All quotes are anonymous unless you wish to be given credit.

8. **Have a study you wish to share?**
   The moderator reserves the right to ask poster for REB approval before recruitment posts are allowed.

Adapted from the Group Rules from 'Parents Partnering in Research' Facebook group.

Sibling Youth Advisory Council (SibYAC)
Facebook Group Rules

1. **Treat people the same as you would face-to-face**
   Remember that there is a person behind the computer screen, and we should remember to treat others with respect in ways that we would in person.

2. **Please do not to share sensitive information**
   The moderator is not responsible for outcomes of postings, so please use your discretion for your postings. If you are unsure of information to post, you are welcome to check in with the moderator.

3. **Spams and engagement with others**
   This is a space to build a community, share information, and connect socially. Please consider the intent of your posts and chat with others. We are your community, so please feel free to say hello!

4. **No medical advice please**
   We encourage open communication and sharing of information. Please note that due to legal concerns, there will be no medical advice given.

5. **Your privacy matters**
   Please remember that this is a safe space for everyone to share their experiences and stories. Nothing that you share (photos, personal information, comments, anecdotes) may be repeated or used by others in another setting or publicly without your expressed consent. If you are asked for permission, you are not obligated to agree to it. It is entirely up to you what you decide to share.

6. **Research information**
   For information related to Linda Nguyen's doctoral studies, please remember that the research is being conducted and refrain from sharing the research with others before having a discussion in this group.

7. **Data collection may happen on occasion**
   Information in this group may be used for data collection and/or publication purposes. This is never done without your permission. All quotes are anonymous unless you wish to be given credit.

8. **Have a study you wish to share?**
   The moderator reserves the right to ask poster for REB approval before recruitment posts are allowed.

Adapted from the Group Rules from 'Parents Partnering in Research' Facebook group.

Supplementary File 4. Structure of team meetings with the Sibling Youth Advisory Council.

The SibYAC requested that they like to have monthly team meetings, which provided an opportunity to connect as a group. The meetings are scheduled for 1-hour based on the availability of the SibYAC. An online link using the Zoom software platform with a toll-free number was provided to ensure that the meetings were as accessible as possible. The first half of the meeting was a check-in, in which there are informal conversations about how each member felt in their day-to-day lives. The second half of the meeting focused on providing updates, feedback, and comments on the research projects. The end of each meeting concluded with an invitation for the SibYAC to be involved in completing follow-up action items related to the research projects. These items could be completed by email or in a separate small group meeting.

Supplementary 5. Funding support for the SibYAC partnership

To acknowledge stakeholders contributions of their expertise, compensation for their time involved in collaborative and consultative activities is important to consider (149). After a year of partnering in my PhD studies, I applied and was successful in receiving two fellowship awards with the support of the SibYAC. The SibYAC reviewed the grant applications and provided testimonials about the importance of the proposed research projects. First, I received the CIHR Fellowship: Patient-oriented Research Award – Transition to Leadership Stream – Phase 1 that provides a research stipend of $50,000 and a research allowance of $5,000. Second, I received the CHILD-BRIGHT Graduate Fellowship in Patient-Oriented Research of $9,990 to support one of my doctoral projects and partnership with the SibYAC. Both fellowship awards allow me to provide compensation to the SibYAC based on the guidelines in the Terms of Reference (Supplementary File 2).

CHAPTER TWO

TITLE: Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis

AUTHORS: Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, Ketelaar M, & Gorter JW

JOURNAL: Frontiers in Rehabilitation Sciences, Families and Functioning in Childhood and Adolescence issue

CITATION: Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Frontiers in Rehabilitation Sciences. 2021:52. doi: 10.3389/fresc.2021.724589

Abstract

Background: As children and adolescents with a chronic health condition (CHC) age and transition to adulthood, many will increasingly assume responsibilities for the management of their healthcare. For individuals with CHCs, family members including siblings often provide significant and varied supports. There are a range of resources in Canada to support siblings of individuals with a CHC, but these resources are not synthesized and the extent to which they relate to healthcare management remains unclear.

Purpose: The purpose of this document review was to identify, describe, and synthesize the types of resources currently available to provide general information and healthcare management information about how siblings can provide support to individuals with CHCs in Canada.

Methods: Print and electronic resources were systematically identified and retrieved from the websites of organizations, treatment centres, and children's hospitals that are part of Children's Healthcare Canada. Each unique resource was treated as a text document. Documents that met the following inclusion criteria were included: addressed the topic of siblings of individuals with a CHC and written in English. Data were extracted from included documents and qualitative conventional content analysis was conducted. Throughout the process of this review, we partnered with a Sibling Youth Advisory Council.

Results: The systematic search yielded 1628 non-duplicate documents, of which 163 documents met the inclusion criteria. Of the total of 163 documents, they were delivered in the following formats: 17 (10%) general informational products (e.g., booklets, videos) about a CHC and sibling relationships, 39 about support programs and workshops (24%), 34 news articles (21%) that described the roles of siblings, and 6 (3%) healthcare management informational products (e.g., toolkit, tipsheets), 31 blogs (19%) and 39 interviews (24%) with parents and siblings. In the blogs and interviews, siblings and parents described how siblings developed knowledge and skills for healthcare management, as well as their role and identity over time.

Significance: This study identified that there are limited resources available about healthcare management for siblings of CHC in Canada. Resources are needed to facilitate conversations in the family about the role of siblings with healthcare management of their sibling with a CHC.

Introduction

In North America, approximately 15% to 18% of all youths have a chronic health condition (CHC) (1,2). The term "chronic health condition" encompasses congenital and acquired diseases, as well as physical and mental health conditions (3). There has been a shift towards providing care for a family of a child with a CHC using a non-diagnostic approach (4). Instead of focusing on a specific diagnosis or condition alone, increasing care and supports focus on providing comprehensive care to address the holistic needs of the individual and family (4,5).

Families often express significant concerns about how they can best support their child during the transition from the pediatric to adult healthcare systems (6). During this time, youth with CHCs will need to learn how to manage their healthcare, for example learning how to navigate the process of filling prescriptions, scheduling healthcare appointments, and answering questions from healthcare providers. Typically, support is provided by family members through this transition period. In addition to healthcare management, individuals with CHCs are also exploring their interests and goals, including school, work, and leisure (6–9) and learning to navigate new environments, including healthcare for adults, education, transportation, recreation, and social services (7). Throughout the lifespan, families typically can provide support, given their past experiences coordinating their family member's or child's care and knowledge of their child's strengths, areas of improvement and goals (10).

In addition to parents, siblings are a part of the family who can provide support for their sibling with a CHC. Within the typical lifelong bonds between siblings, these relationships are highly dynamic and can change over time depending on the needs, roles, and commitments of the whole family (11–13). Each sibling relationship is different with varying levels of emotional closeness, social connectedness, and expectations of each other (14). During childhood and adolescence, sibling relationships are unique as they often live and grow up in their shared home environment where they can act as peers, confidants, or role models (15). At a young age, siblings of individuals with a CHC often recognize that they need to support their family in different contexts (16,17). When there is future planning involved from the whole family, siblings often feel closer to each other and with their family, and they have a clearer understanding about their role for the future during adulthood (18,19).

In some families, there may not be discussions about the role of siblings but siblings may be expected to become carers to their sibling with a CHC (20). In 2018, the Siblings Needs Assessment Survey was conducted in Canada among young adults who were ages 20 years or older, who had a sibling with a disability and received a total of 360 responses (21,22). Siblings described concerns for their sibling's future such as finding employment or living independently (12,23). Siblings might have worries for new responsibilities, such as guardianship or financial responsibilities, when their parents can no longer be the primary caregivers (13,24,25). These concerns can affect the extent to which siblings are involved in the healthcare of their sibling with a CHC.

Typically developing siblings might want to support their sibling with a disability, but they require knowledge and skills on how to do this. There are currently 'Sibshops' that are offered across ten countries, including the United States and Canada (26). These Sibshops provide an opportunity for siblings to connect with people with similar experiences and share stories. A survey was conducted to evaluate Sibshops, in which 66% of respondents identified that they learned coping strategies, 75% reported that Sibshops had a positive impact on their adult lives, and 94% stated that they would recommend Sibshops to others (27). Often, one of the goals of Sibshops is to provide a space for siblings to meet and share experiences with other siblings of individuals with a CHC in a recreational setting (28). Some SibTeen sessions are also held for adolescents ages 13-17 years old to offer a community of support (29). Although there are support groups for siblings of individuals with a CHC, such as Sibshops and SibTeen sessions, there are no tailored resources or programs for typically developing siblings to share their concerns about supporting their sibling with a CHC specifically for healthcare management. There are many ways that siblings can provide support to their sibling with a CHC. These can be categorized as: concrete support such as taking on responsibilities and providing assistance; emotional support such as listening and empathizing; advice support such as offering information; and esteem support, such as expressing encouragement (30). Siblings can offer these different types of supports to help their sibling with a CHC manage their healthcare.

Informational needs have been identified to be a critical need for siblings (31). Siblings who wish to have a caregiving role often seek knowledge in how to provide care to their sibling with a CHC, how to navigate disability services, and how to seek supports for themselves (31). While there is information available and advertisements of services for siblings on websites of children’s hospitals and organizations, many families and siblings identified that they were not aware of this information (32,33). Siblings identified that they want to have an open, constructive dialogue with their parents about the future, including expectations and responsibilities (13,24). Often, siblings had to learn how to care for their sibling on their own as information was not always passed down from parents to the siblings (11,24).

Informational needs are also increasingly being addressed by individuals, including siblings and their families, through the use of the Internet. Siblings can share their experiences and needs online in various formats, such as blogs. Among the few studies that have analyzed the content of blogs, researchers identified how individuals who write these blogs can share experiences that might be different from what might be shared in a research study. Young adults and families have previously written blogs to document their experiences in healthcare, including their emotions and challenges (34–36). Similarly, blogs written by siblings and families can provide insights into the needs of siblings in order to prepare for their roles with healthcare management. There is a gap with little information known about the types of needs about healthcare management that siblings of individuals with CHC are sharing online.

Individuals may also choose to find information online for various reasons, including medical information, such as options for therapy, treatments and health services (37,38). Siblings of individuals with a CHC require information on how they can provide support with healthcare management. In the Canadian Siblings Needs Assessment Survey conducted in Canada, the majority of respondents across all age groups identified online websites as their preferred method for resources, information and tools (22). Programs, such as Sibshops and SibTeen sessions in North America, are often promoted online and share information about eligibility criteria and registration. In other countries, initiatives to support the needs of siblings of individuals with a CHC include Siblings Australia developed in 1999 (39), Sibs in the United Kingdom in 2001

(40), and the Sibling Leadership Network in the United States in 2007 (41). These initiatives in Australia, the United Kingdom, and the United States have been established for many years, and includes an array of support programs and resources for siblings of individuals with a CHC. In Canada, the Sibling Collaborative was established in 2017 and offers online support groups with some resources such as information about the COVID-19 vaccine, finances, and stories from siblings (42). Despite the availability of many resources, as a team, we have heard from siblings that resources about healthcare are not easily accessible or retrievable in Canada. The resources are often posted on certain websites by children's hospitals and organizations, but the websites are not easy to navigate. In Canada, there is no national systems approach to store resources for siblings of individuals with a CHC. Considering the important and multi-faceted roles that siblings can have, it is important to identify and summarize the different types of resources that are available to siblings of individuals with a CHC.

This review aims to identify and describe:

i. the types of resources currently available in Canada to provide both general information and specific healthcare management information about how siblings can provide support to their sibling with a CHC; and
ii. key topics discussed in resources created by siblings and families.

Methods

Integrated knowledge translation

An integrated knowledge translation approach was used throughout the process of this review to partner with the Sibling Youth Advisory Council (SibYAC) comprised of six young adults who have a sibling with a disability. The SibYAC were first involved with the idea and concept, as well as the research question of this review. The SibYAC shared their experiences with searching for information to support their roles as siblings, and they identified a need to identify and synthesize resources that are available to siblings of individuals with a CHC. These experiences from the SibYAC provided a clear rationale to support our review aims. There were individual check-in meetings with each SibYAC member, and an engagement framework (43) and Involvement Matrix (44) were used as tools to ask about the tasks and roles that they would like to have in this review. The SibYAC were further involved in data analysis by sharing their perspectives for the retrieved documents to ensure that the extracted data are synthesized meaningfully for siblings, families, and other stakeholders. They were then involved with the interpretation of results and drawing conclusions. Meetings were held with the SibYAC to ask about their reflections of the summary of results with guiding questions including: How do the documents and websites support siblings in their role? Based on the documents and websites, what are some needs, information, or questions that you still have as a sibling? For example, in healthcare or in general. Reflections from the SibYAC helped to identify the gaps and future directions about resources to support siblings in their roles, including with healthcare management of their sibling with a CHC.

Qualitative document analysis

Qualitative document analysis involves a systematic search of documents and resources, which includes both printed and electronic resources (45). A variety of documents can be analysed, including books, brochures, diaries, journals, event programs, or news articles.

Search strategy

A comprehensive search was conducted on publicly available websites of thirty-one organizations, including children's hospitals, and rehabilitation centres that are part of Children's Healthcare Canada (46). These were selected to provide an initial understanding about the types of resources that are available for siblings of individuals with a CHC in a healthcare setting. The websites were searched in August 2020. A broad search strategy was employed in the search engine of each website with the terms: "sibling", "brother", or "sister". All documents from the search were digitally retrieved using a feature called NVivo Capture and imported into NVivo (Version 11.4.3). Duplicates of documents across websites were removed.

Inclusion and exclusion criteria

Text from all retrieved documents was initially scanned in NVivo for the key terms of 'sibling', 'brother', or 'sister'. Documents that included at least one of these key terms were read by the first author (LN). Identified documents were included in the review if they: 1) addressed the topic of relationships between siblings with and without a CHC; and 2) were published in English. Documents were excluded if the sibling was mentioned but did not discuss supports of siblings or the relationship between siblings of individuals with a CHC.

Ethical considerations

Ethics approval was not required to retrieve and analyze documents that are publicly available on the Internet. An assessment of online documents can be conducted to identify the intent of online documents and its use in research, and documents that are written for public intent do not require consent from the creators or authors of the documents (47). In the analysis of retrieved documents, there was careful consideration to protect the privacy of the creators for the documents, and all personal identifiers were removed from included documents.

Data extraction and analysis

A data extraction template was created using Microsoft Excel Version 16.41 to collect data from each document (48). This template included the following categories: document source, document type, purpose/goals, and key content. For document types coded as “blog” or “interview,” content data for two additional categories were extracted: 1) family characteristics; and 2) CHC of an individual in the family. Additionally, all blogs and interviews were read and re-read in an iterative process to achieve immersion in the data and understand the stories shared by siblings and parents. Conventional content analysis was conducted by the first author (LN) for documents that were coded as blogs or interviews (49). Initial codes were developed based on the full text of the blogs and interviews, and these codes were then organized into categories to depict how they were related and linked to each other. Codes were grouped into meaningful clusters or categories based on their similarities in concepts. An Excel spreadsheet was created, that included extracted quotes and codes that were grouped into categories. Each category was expanded into a short statement to describe the key topic shared by siblings and families. Two analysis meetings were then held with individuals familiar with the content (e.g., SibYAC) and qualitative analysis (e.g., graduate students, co-author SMJ) to review and name the categories, and identify additional properties and dimensions of each meaningful cluster. Analytic notes were written by the first author (LN) about how the categories related to each other to form meaningful clusters. While the content of all documents was analyzed to identify information and supports for healthcare management of an individual with a CHC, conventional content analysis allows for the identification of key topics from included documents that describe the experiences of siblings of individuals with a CHC beyond healthcare management. In this review, recognizing that gender is non-binary, we refer to siblings as a “brother” or “sister” based on the information provided in the resources included in this review with the recognition that siblings may identify themselves along a spectrum.

Data credibility

To ensure credibility of the data, an audit trail and multiple analyst triangulation were used as two strategies. An audit trial was created to describe the steps and document decisions that were made about data extraction, as well as the identification of codes, categories, meaningful clusters, and key topics identified in the documents (50,51). Sufficient time was also spent reviewing each source of information to identify recurrent patterns and key topics of the documents (52). The first author (LN) spent extensive time to read and re-read all documents, and took field notes of emerging ideas for each document in an Excel document (e.g., What is the main message about this document? How does this document relate to other documents?). To further enhance the credibility and dependability of the data, the lead author engaged in reflexivity and documented their own biases, preferences, and preconceptions about the topic in a series of memos (53,54). Analyst triangulation was employed, in which multiple individuals with different backgrounds and expertise offered their perspectives about the preliminary and final findings (54). Two initial meetings were held to review and discuss how to organize preliminary findings: first with a group of graduate students with expertise in mixed methods and qualitative research, and then with the SibYAC. Two additional meetings were held with the SibYAC to share their reflections about the meaning of the findings in this review to them, describe whether the key topics from the blogs and interviews resonated with, or differed from, their experiences as young adult siblings of individuals with a disability, and identify gaps for future directions. All SibYAC members present at the meeting described that the key topics were similar to their experiences, and they provided suggestions for future directions in the development and enhancement of resources for siblings of individuals with a CHC. All authors of this review are from a multidisciplinary backgrounds including cognitive psychology, education, nursing, occupational therapy, physiatry, rehabilitation, patient-oriented research, and lived experiences, and all provided their perspectives on the synthesis of findings.

Results

The systematic search yielded 1628 non-duplicate documents and resources, with 1015 documents and resources that included keywords of ‘sibling’, ‘brother’, or ‘sister’. There were 163 documents and resources that met the inclusion criteria (See Figure 1).

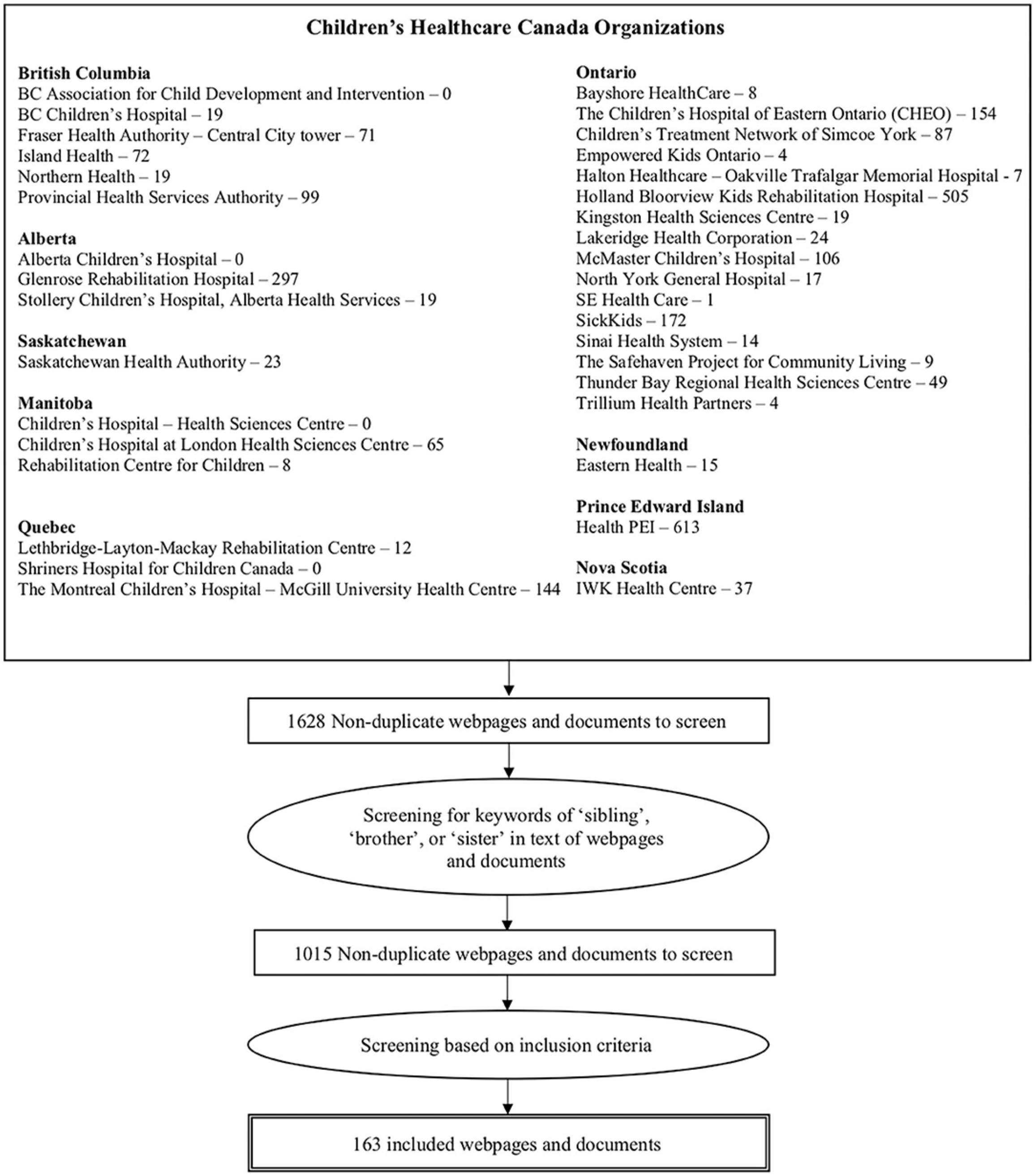

Figure 1. Flowing diagram outlining the section of included websites and documents.

All resources were identified from treatment centres that provide inpatient and outpatient services to children and adolescents with a CHC. Some documents discussed CHCs as a broad group of conditions, while others referred to specific conditions such as autism spectrum disorder, cerebral palsy, Down syndrome, epilepsy, genetic disorders, juvenile arthritis, intellectual disorders, or mental health disorders. The documents included 6 books (55–60), 1 podcast (61), 34 programs and 5 workshops [references provided to websites where ongoing programs (62–67) and workshops (63) are advertised], 7 films and videos (68–74), and 34 news articles. Among the 34 news articles, they referred to 2 announcements (75,76), 6 'awareness' recognition of different days or months of CHCs (77–82), 5 events (82–86), 10 research studies (87–94), 4 participation in research [references provided to websites where active research studies are posted, (95,96)] and 7 stories (97–103). Three documents referenced a toolkit (104–106), 3 booklets (107–109), and 3 tip sheets (110–112). Of the total of 163 documents, there were 31 blogs (19%), of which 13 (8%) written by parents and 18 (11%) written by siblings, and thirty-eight interviews (23%) with 12 (7%) interviews with parents and 26 (16%) interviews with siblings, and one interview with a family that included both the parents and siblings. Table 1 provides details about the document type, source of documents, number of documents and references, target audience, purpose and goals, and summary of key content.

[insert Table 1 about here].

Types of resources for general information

The majority of resources were general informational products for siblings of individuals with a CHC, which are available in a variety of formats. Supplementary Table 1 presents detailed descriptions of these resources.

i. *Booklets and books.* Books were available for siblings and families, in which some highlighted the need to understand the importance of sibling relationships, for example, creating a space for siblings to understand their emotions when they have a sibling with a CHC. Booklets were also available to provide guidance to parents and teachers about how to communicate with siblings of someone with a CHC.

ii. Podcasts. Personal stories from families, including siblings, were shared through podcasts, films, and videos. These stories described the journey of the whole family, and one podcast discussed the relationship between the siblings in which one sibling has a CHC.

iii. Programs and workshops. There are advertisements that announced past programs (n=34) and workshops (n=5) available to siblings and families. Among these 39 documents, 17 were in-person, 8 were virtual due to COVID-19, and 14 did not indicate the type of format. Most programs offered were sibling support groups, such as Sibshops, that are available throughout the year for siblings who are ages 7 to 25 years old. There were programs specifically for families to connect with other families of children with autism available for free [reference to ongoing advertisement about the program, (57)].

iv. News articles. All stories that were published as a news article were authored either by parents (n=1), mothers (n=2) or a sibling (n=3), both a mother and sibling (n=1). Articles authored by mothers focused on stories of their child's lived experience, parenting multiple children with a CHC or the same CHC, and/or the roles that other children may assume when there is a child with a CHC. Siblings discussed topics, such as sharing their emotions about their sibling relationship, providing support with healthcare management, and transitioning into different roles as a sibling, such as becoming a caregiver. Throughout the year, there were news articles with announcements about initiatives that were inspired by the stories of siblings. For example, there were announcements about various 'awareness' days and months about specific disabilities and health conditions, which provided an opportunity for siblings to share stories about their sibling with a CHC (69–74). News articles also advertised research studies that were completed or actively recruiting sibling participants. The topics of these studies included genetic studies for specific health conditions, such as autism spectrum disorder and lymphoma, successful organ transplants between siblings, effectiveness of assistive equipment, and a survey to understand the needs and feelings of siblings of youth with a CHC.

Type of resources for how siblings can provide support with healthcare management

There are few resources that provided information for siblings about their roles with respect to the healthcare management of their sibling with a CHC. When resources were available, they were formatted as either tip sheets or as a toolkit.

Toolkit and tip sheets

Both parents and siblings could refer to different sheets that were available for download online, which included tip sheets (110–112), and a toolkit (104). These sheets also focused on strategies for how siblings can provide support to their sibling with a CHC. For example, there was a tip sheet that described strategies for siblings of inpatients at a children's hospital (112). Some strategies for how siblings can be included as part of the inpatient stay were being a part of their sibling's care team, doing fun activities together, talking to their sibling, and helping the sibling to decorate their room (112). While the resources primarily focused on providing knowledge about a CHC to siblings, some resources provided additional strategies for siblings to support the healthcare management of their sibling with a CHC. A toolkit was also co-designed with siblings, clients, parents and clinical staff for brothers and sisters of children who have an acquired brain injury (104). The toolkit was described as a resource that siblings can use to learn knowledge about their sibling with an acquired brain injury and learn how to explain this injury to other adults who can provide help, when needed (104).

Blogs and interviews

Siblings and families described different types of CHCs in blogs and interviews. Some siblings also had the same CHC as other siblings in the family. The age of siblings and individual with a CHC discussed in blogs and interviews ranged from infancy to older adults. The size of families ranged from one to five children. Based on an analysis of the content from blogs and interviews shared by siblings and parents, a conceptual map was developed to describe the codes, categories, and key topics discussed in these documents (See Figure 2). Detailed descriptions about these key topics are described in detail below. The frequency that these topics were identified in the blogs and interviews are provided in Table 2.

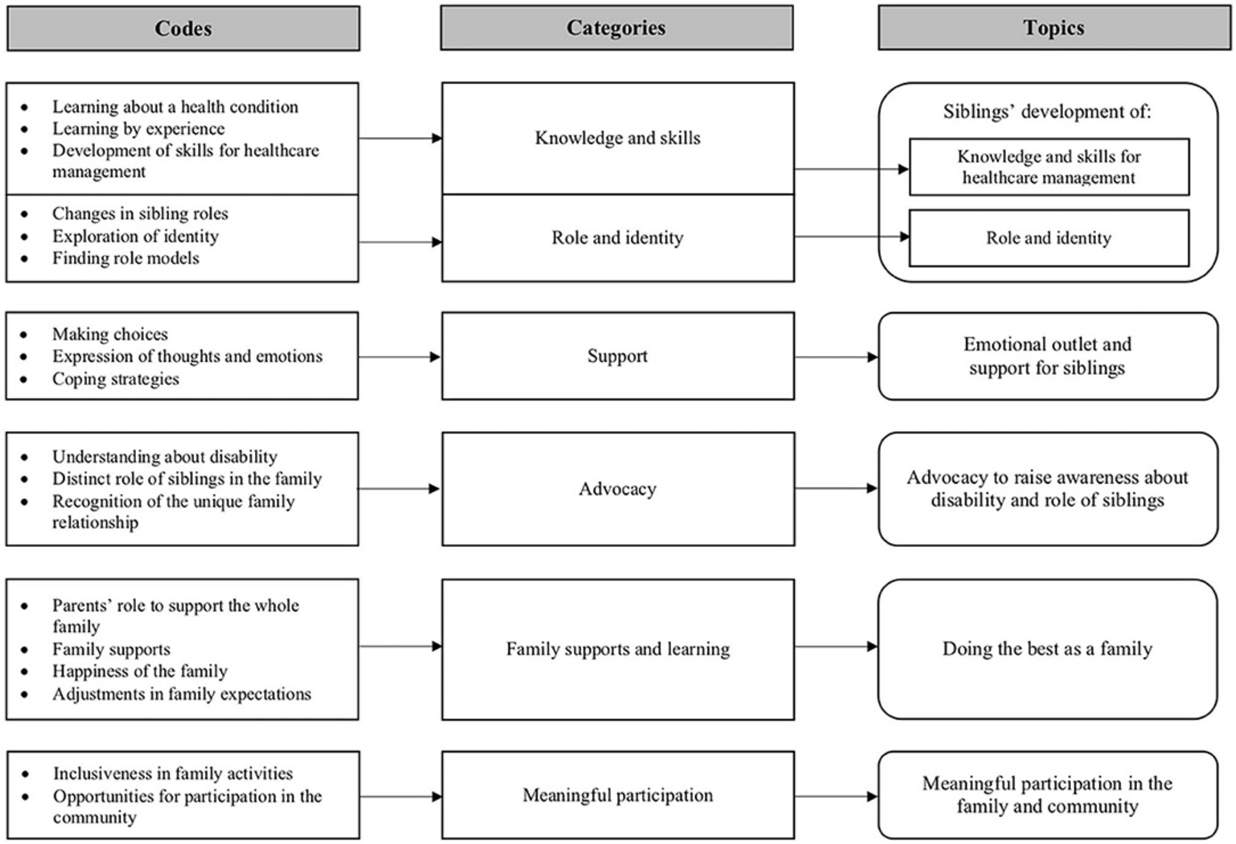

Figure 2. Codes, categories, and topics identified in the blogs and interviews.

Table 2. Frequency of topics identified in interviews and blogs with parents and siblings.

Topics		**Siblings**		**Parents**	
		Blogs (*n* = 18)	**Interviews** (*n* = 26)	**Blogs** (*n* = 13)	**Interviews** (*n* = 12)
Siblings' development of:	a) knowledge and skills for healthcare management	7	8	4	5
	b) role and identity	12	16	4	6
Emotional outlet and support for siblings		7	12	–	3
Advocacy to raise awareness about disability and role of siblings		6	3	–	–
Doing the best as a family		–	2	5	6
Meaningful participation in the family and community		6	4	–	5

–Topic was not identified.
** Multiple topics may be identified in each interview and blog.*

Siblings' development of knowledge and skills for healthcare management

Siblings described how they needed to learn about the meaning of disability. Some siblings did not understand specific CHCs, such as the different treatments and services their siblings had to receive to manage their CHC. Siblings often described that they simply saw their sibling for who they were, regardless of the CHC. Parents shared stories in their blogs about the forming of relationships between their children. Young children learned how to develop their relationship with their sibling with a CHC. A mother shared the story of how she saw her two children interact with each other, where her young daughter asked to hug her brother or hold his hand when he was using an assistive device to walk. As siblings began to develop an understanding about the CHC, some siblings offered support with healthcare management. For example, a mother described how her daughter learned to be present and hold her brother's hand when he was using a suction machine. Siblings shared in interviews about how they learned different ways to support their siblings. For example, a sister observed her mother apply breathing techniques with her brother and she learned how to do the same. There was a process in which siblings first needed to learn about the CHC and develop a relationship with their sibling with a CHC, which then allowed them to learn how to offer support with healthcare management.

Siblings' development of role and identity

The role of being a sibling to someone with a CHC provided them with experiences about a CHC, and the sibling role became a part of their identity. Young adult siblings shared in written blogs about how they were developing their own identity, such as moving away for university and developing their career. For some siblings, the experience of growing up in a family of an individual with a CHC motivated them to pursue a career to support other children with a CHC, such as healthcare professions and research about a CHC. Siblings would bring their personal experiences about a CHC into their professions, such as an understanding about a CHC in research or how to interact with families. Their personal experiences about a CHC also motivated them to use their academic knowledge to create resources, such as mobile applications or tools that children with a CHC could use. Both parents and siblings identified multiple roles that siblings had in the family. Siblings continued to maintain a close relationship, and when one sibling had an acquired CHC, the siblings would learn how to provide support to each other. For example, a sibling described how he went to therapy appointments with his brother who had an acquired brain injury. The sibling provided both support and humour by being present at the therapy appointments, and the parents described how the sibling became a part of the care team. Adult siblings described challenges that they had when they became a caregiver, such as the sacrifices that they had to make with living with their sibling with a CHC or not being able to work full-time.

Emotional outlet and support for siblings

Siblings wrote in blogs that they shared with the community about both the positive aspects of their relationship with their sibling with a CHC as well as the challenges. Siblings shared the message about how they are not alone, where their thoughts and feelings matter. Some siblings pursued their own goals and happiness, and also chose their roles with their sibling with a CHC. Some adult siblings experienced guilt when they did not voice their opinions. One adult sibling shared her sense of guilt when her brother was sent to an institution that the family believed was a good option at the time. Siblings spoke about how their emotions were connected to the emotions that their parents were experiencing, such as frustrations and stresses. Some siblings wanted to find ways to address the challenges such as learning how to help their sibling with a CHC. With the different emotions that siblings were feeling, they sought ways to have an outlet to express their emotions. A Photovoice program was offered to siblings and young patients with cancer, where they took photographs that represented their experiences that were later displayed at an event for the public community. Siblings were developing skills in how to cope with their emotions, and parents identified how these siblings would develop personal skills such as being caring and empathetic. Siblings also shared about the importance of open communication with parents because siblings might have hidden emotions. Siblings identified that they might not initiate discussions with parents about their feelings, and parents can create a space for these discussions.

Advocacy to raise awareness about disability and role of siblings

Some siblings became advocates, in which they expressed a need to explain what disability was to their peers. For example, a sibling of a brother with autism described how she read a book to her class to explain autism and she often took the time to answer questions from her classmates. Siblings valued the connection that they had with other siblings who had similar experiences. Some siblings grew up without knowing about other siblings who have a sibling with a CHC. Siblings wanted to connect to a community of siblings to not only advocate for CHC and disability awareness and supports, but also learn about the role that siblings can have. For example, some siblings did not realize that they developed skills that could be well-suited for healthcare professions. One sibling described how she learned about the profession of a child health specialist after connecting with another sibling. Furthermore, adult siblings who were caregivers or guardians of their sibling with a CHC they identified how their roles were often not recognized at work. For example, employers recognized when co-workers needed to leave to take care of their child but not for their sibling with a CHC. Siblings identified how there should be recognition of the important role that they have. They all wanted to be part of a community where they can create change and advocate for a diverse community that their own sibling with a CHC could meaningfully participate in.

Meaningful participation in the family and community

Families identified the importance of creating an inclusive environment where a child with a CHC can participate in activities. Some families planned trips and made sure that they rented adaptive equipment to ensure that their child with a CHC could participate in activities, such as hiking, biking, or kayaking. In their daily lives, young siblings shared in their blogs that they made sure that their sibling with a CHC was included in the games that they played with their friends. One family thought about different ways that every member of the family could participate in activities. When a sibling might be attending speech therapy, other family members could coordinate to have the other siblings participate in a sports activity at the same time. Parents identified how it was important to make sure that all siblings could meaningfully participate in the community. Both parents and adult siblings expressed their concerns about opportunities for their sibling with a CHC to participate in the community in the future. Siblings shared the positive value of a job for their sibling with a CHC, which provided a sense of pride to participate in the community. Some siblings wanted to address concerns about how to create an inclusive community for people with CHCs, and they created mobile applications to encourage their sibling with a CHC to develop the skills needed to participate in the community. For example, one sibling created a mobile application with a set of cards with which an individual with autism could practice the skills they needed to carry out an activity, such as taking public transportation. Both parents and siblings sought opportunities for a sibling with a CHC to participate in the community as they grow older.

Doing the best as a family

During separate interviews, parents shared about how they were doing the best that they could as a family and siblings shared how the journey of every family was different. A mother shared in her blog about experiences with raising her children, including children with a learning disability and Down syndrome, and she needed to time to learn about her children. For other parents, they learned about the different types of supports that would be appropriate for their child with a CHC and there was no ‘one size fits all’ approach. Some parents initially chose to keep their life private, and they did not want to burden others with the responsibilities in caring for their child with a CHC that they feel were their own. They gradually recognized how it was important to reach out to others for support, such as their children and neighbours. Some parents also sought respite services to take care of their own health in order to optimize the care that they could provide to all of their children. In addition to services, parents described the value of building a network of supports, such as connecting with other families with similar experiences. They wanted to have opportunities to meet other families and participate in activities that included the whole family. Some families created videos and films to share their story of both the positive experiences and challenges with other families.

SibYAC reflections on the findings

After synthesizing the findings from this review, the SibYAC members were asked to share their perspectives about the meaning of these findings. There were key topics raised in the blogs and interviews included in this review, and the SibYAC members were asked about whether these topics resonated with their own experiences of siblings of individuals with a disability. Siblings who wrote the blogs and interviews included in this review identified that they wrote blogs as a way to share their stories so that other siblings would know that they are not alone, and writing blogs was an outlet for their emotions. Similarly, SibYAC members also wrote personal blogs about their personal experiences as a sibling and the roles that they have had. One SibYAC member shared an excerpt of her journal while her brother, who has cerebral palsy, was in a rehabilitation hospital after orthopedic surgery: “As my brother began to see progress into the next day, so did I. As he found a rhythm and learned the shuffles of the hallway, so did I. And before I knew it, I fell head over heels into the routine of physical and psychological exhaustion but unimaginable emotional fulfillment.” She shares that her personal experience is a clear example of why consciously integrating siblings into the family-centred care model is so important.

While the findings of this review help to identify key resources for general information and information of how siblings can provide support with healthcare management, the SibYAC continued to identify that there is a need for advocacy to raise awareness about the important roles that siblings have. They often had to learn to develop knowledge and skills, in order to have a role with supporting their sibling with a disability with healthcare management. A SibYAC member shared: “There is no handbook for special needs siblings. It's not something that's majorly talked about and kind of always felt like a big secret. Every day, I am learning more about how to appropriately support my sibling through the transition from pediatric into adult healthcare.” While this review identified that there are resources available for siblings of individuals with a CHC, few resources offer support for how siblings can be involved with the healthcare management of their sibling with a CHC.

As the SibYAC reflected on these findings, there is a critical gap in which there are no online resources available from Children's Healthcare Canada to support siblings in conversations about healthcare management and future planning to their sibling with a CHC, even though many siblings might already be part of the care team. A SibYAC member shared her personal experiences with this gap: "There has never been planning about the present, day-to-day things, let alone future planning, that has included me. The extent that I have been involved with my siblings is equal to the amount of intention and force I used to create a space for myself." The SibYAC shared how discussions about future planning can be helpful for families to ensure that there is clear communication about the role of siblings.

Discussion

This review identified a variety of resources and documents available in English for siblings and families of children with CHCs across organizations of Children's Healthcare Canada. Most resources consisted of general information for siblings and families: to become aware and learn about different CHCs through books, news articles, and podcasts. There is an increasing trend in the use of the Internet for health information among patients, their families and general public (113), and each family requires different types of information based on their needs (114). In a qualitative study to explore the experiences of parents of children with disabilities who sought information, parents used online information to supplement the information provided by professionals (38). When healthcare professionals did not provide enough information during a consultation, some parents searched on websites of hospitals and rehabilitation centres for additional medical information (38). In the search for information, parents may also identify resources for how they can support the siblings of a child with a CHC. This review retrieved booklets for parents about how to communicate with siblings of children of individuals with a CHC.

In this review, few resources were identified to support siblings in the role of healthcare management with their sibling with a CHC. Similar to parents, siblings might also have questions and would like to have more information to support their roles in the healthcare management of their sibling with a CHC. Siblings of individuals with a CHC require skills and knowledge if they choose to have an active role in the healthcare management their sibling with a CHC. There are tip sheets that provided guidance for siblings to build a relationship with their sibling with a CHC, such as playing games or doing activities, as well as how to be a part of the care team (111,112). There is also a toolkit that originally developed for siblings of individuals with an acquired brain injury, recently expanded to include different disabilities (105). It is important to consider and tailor different resources to prepare for the roles that siblings might choose to have, including with healthcare management of their sibling with a CHC.

This review highlighted a key gap in the needs of siblings based on the personal stories that they shared through blogs; many identified a need for emotional support. Blogs can be helpful for siblings, where they might find comfort to know that they are not alone (115). Siblings require acknowledgement of their emotions, and for some siblings, they are learning how to address their emotions as they continue to have a role in healthcare to their sibling with a CHC. Siblings might choose to seek online support to be part of a community with others who have similar experiences (115). Parents and families have previously described the importance of being a part of an online community where they can seek resources and connect with other families (38,116), and siblings might have similar motivations to connect with other siblings online.

Siblings also shared, in blogs and interviews, their perspectives about the importance of advocacy to raise awareness about CHCs and their own roles. Siblings may need to advocate for their role in the family. This review identified that siblings require additional resources in order to learn and be prepared for their future roles. The extent of discussions about future planning can vary in families, and siblings are often not included in these discussions (13,117). While some parents may wish for the siblings to have their own separate lives, siblings shared in qualitative studies that they chose to have active roles such as being a caregiver and they identified the need to have conversations with their parents about future planning (118,119). Discussions about future planning can be helpful for families, providing an opportunity for siblings to identify new or changing roles and to facilitate the sharing of information between parents and siblings (31,33). These discussions can be ongoing to adapt to the changing situations of the family over time. While these discussions can be challenging and complex for families, siblings identified the need to have clear plans so that they can be prepared for their future roles (117,119,120).

Many families shared how they are learning from experience and doing the best that they can their family member with a CHC. In this review, both parents and siblings described the positive value of creating a supportive and inclusive environment of a person with a CHC. This inclusive environment applied to the family environment where some families sought opportunities to participate in different activities, as well as in the community such as having a job. Both parents and siblings described the concern that they had for an individual with a CHC after graduating from high school, and they were worried that there may be fewer opportunities to participate in the community. This concern about the transition to adulthood for individuals with a CHC has been raised in the literature (121,122), and current news noting that there are approximately 12,000 young and middle-age adults with disabilities in Ontario, Canada who are on a waiting list to seek supports and residential care (123). In addition to employment opportunities, both parents and siblings have future worries for their child with a CHC such as the navigation from pediatric to adult healthcare services (122,124). Some siblings shared in blogs and interviews how they gradually learned to take on caregiving responsibilities. Siblings are becoming adults and they may take on future caregiving responsibilities. They are often learning through experience about how to care for their sibling with a CHC throughout the lifespan (119).

Strengths and limitations

A strength of this review is the involvement of the SibYAC as advisors throughout the process of this review. They provided their perspectives on the aims of the review, data analysis of resources, and future directions on how to disseminate the findings and develop future resources. Another strength of this review is that the resources identified have been compiled and can be applied to enhance existing resources for siblings of individuals with a CHC and inform the co-development of future resources. A limitation of this review is that the information that can be extracted from the documents included may be restricted by the purpose of the document and the content that the creators choose to share. Another limitation is that all documents were identified from children's hospitals and treatment centres that are part of Children's Healthcare Canada were in English and excluded documents in French. The documents and resources from the websites of organizations that are part of Children's Healthcare Canada might be selectively published may not include information about the challenges of the care that these organizations provide. The information may not be reflective of the entire landscape of resources for Canadian siblings of individuals with a CHC. In addition, at the time of this review, documents and resources were retrieved from 31 organizations that were a part of Children's Healthcare Canada. Since then, eight additional organizations have been included. Most organizations that are part of Children's Healthcare Canada are children's hospitals and rehabilitation centres, and resources from services offered in the community, such as mental health services, might have been missed. However, the resources and documents in this review provided a starting point for identifying general information and information about how siblings can support their sibling with healthcare management. Additionally, while data extraction and coding was conducted by a single analyst which limits our ability to report on inter-coder reliability, the categories and meaningful clusters of data that were developed were reviewed and discussed by two key stakeholder groups, a form of analyst triangulation and peer debriefing that enhances overall data credibility.

Future directions

This review highlighted key gaps that can be addressed in the future in order to optimize supports for siblings of individuals with a CHC. First, access to existing resources for siblings can be improved by compiling and storing them in one place. As knowledge translation and dissemination can include multiple strategies, there can be multiple formats of resources created, such as infographics, toolkits, videos or podcasts. The Health Hub in Transition in Canada (125) and the F-words for Child Development Knowledge Hub (126) are examples of where information and tools are available online. The uptake and impact of the knowledge translation and dissemination strategies can be evaluated. Second, resources can be offered to support siblings in the healthcare management of their sibling with a CHC. In this review, both parents and siblings shared in blogs and interviews the important role of siblings in healthcare management. Despite the important role that siblings might want to have with healthcare management, there are few resources available to support and empower siblings in this role. Third, this review identified that there are no resources in English available within the online materials from the organizations through Children's Healthcare Canada for parents or siblings to facilitate ongoing conversations about the roles that siblings would like to have with their sibling with a CHC. The conversations could also include the topic of healthcare transition about how youth, siblings, families, and healthcare professionals can help youth prepare for the transfer to adult healthcare (127). Tools could be developed to facilitate these discussions in the family and with healthcare professionals (32,118). Finally, resources could be developed for other professionals, including teachers and healthcare providers, to encourage discussions about the experiences and roles of siblings beyond healthcare management. Siblings have identified that they wanted more information about future responsibilities, such as legal and financial information regarding the care of their sibling with a CHC (22,24).

Conclusion

This review identified resources for siblings that are available from children's hospitals and organizations that are part of Children's Healthcare Canada. Resources that are available for siblings of individuals with a CHC mainly address general information, such as support programs and workshops. There are some resources, such as tip sheets and a toolkit, to offer strategies for siblings to learn about the healthcare management of their sibling with a CHC but these resources are only available at two children's hospitals. Siblings shared about their experiences in blogs and interviews, including their development of knowledge and skills for healthcare management, as well as roles and identity that often relate to the healthcare management of their sibling with a CHC. There is a key gap in available resources, in which siblings and parents identified that knowledge and skills for healthcare management is an important role for siblings but there are few resources that provide this information. Given the needs expressed by siblings, future resources could be developed to share information about healthcare management for siblings, as well as tools to facilitate family discussions about the roles that siblings would like to have in the future. A synthesis of the identified resources could be shared in an accessible format, such as in an online hub, for siblings and families.

Conflict of Interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Author Contributions

LN, BDR, SMJ, MK, and JWG contributed to the conceptualization and design of this review, and perspectives from HD, SB, and HD informed the review aims. LN drafted the manuscript; HD, SB, and JH contributed to the draft of the 'Reflections' section. All authors provided input for the analysis and interpretation of findings. All authors were involved in the revisions, reviewed the manuscript and provided their final approval.

Funding

This review is supported by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream – Phase 1 held by LN. The partnership with the Sibling Youth Advisory Council was financially supported by the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network held by LN. During the work presented in this article, the Scotiabank Chair in Child Health Research was held by JWG.

Acknowledgments

We gratefully acknowledge our partnership with the Sibling Youth Advisory Council throughout all phases of the review. We would like to thank Peter Rosenbaum for his constructive feedback and edits of the manuscript draft.

Table 1. Description of documents.

Document Type	Source of Documents	Number of Documents and References	Target Audience for Resource	Purpose and Goals	Summary of Key Content
Resources for Healthcare Management Information					
Toolkit	One hospital in Ontario	3 (1–3)	Siblings of an individual with a CHC	To offer strategies for siblings of individuals with an acquired brain injury	All documents referred to one toolkit, the SibKit 1.0 (2), which provides information about strategies for siblings of individuals with an acquired brain injury.
Tip sheets	One hospital in Ontario and one hospital in British Columbia.	3 (4–6)	Parents and siblings of an individual with a CHC.	To offer strategies for parents to support siblings, and for siblings to support their sibling with a CHC.	One tip sheet described how parents can support siblings of an individual with a CHC. Two tip sheets offered strategies for siblings to cope with the surgery of their brother or sister, as well as, when their brother or sister is an inpatient.
Resources about General Information					
Booklets	One hospital in Ontario and one service provider in Prince Edward Island.	3 (7–9)	Parents and educators of an individual with a CHC.	To offer strategies for parents and educators of an individual with a CHC and their siblings.	Two booklets, one for parents and one for educators of individuals with autism spectrum disorder, included a booklist relevant for siblings to learn about autism. One booklet offered strategies for educators to support siblings of individuals with childhood cancer.
Books	Two hospitals in Ontario.	6 (10–15)	Public	To share about the importance of relationships of siblings, when a sibling has a CHC.	The books recognize how there is a needed space and role of siblings.
Podcast	One hospital in Ontario.	1 (16)	Public	To tell the stories of people with a CHC, including the experiences of siblings in families with a child with a CHC.	One podcast spoke about the stories of siblings of people with a CHC.

Programs	Four hospitals and two service providers in Ontario.	34* (17–22)	Parents/caregivers and siblings of an individual with a CHC.	The purposes of the programs included providing opportunities for families, including siblings, to connect with other families of individuals with a CHC, spend together as a family through various community events, providing support to siblings to understand the CHC and develop coping strategies.	Programs included: • Family Nights held on a regular basis (e.g., monthly) for siblings and families of individuals with a CHC to connect with each other. • Informal family playgroups that have regular events (e.g., monthly) for families to participate in the community, e.g., indoor playground, escape room, arts and crafts, board games. • Support program, including the Sibshops with sessions for siblings with a range of ages from 7-25 year olds to support siblings who may have questions or are seeking coping strategies with their brother or sister with a CHC. • Bravery Beads Program that allows children an opportunity to collect a different bead for each procedure or event while visiting the hospital for a treatment. An example of how the beads were used between siblings, where the sister educated the class on what her brother went through and still has to go through.

Workshops (one-time event)	One hospital in Ontario.	5* (18)	Siblings and parents of a child with a CHC	The purposes of the workshops were to provide opportunities for siblings to connect with other siblings, discuss ways that parents can support siblings of individuals with a CHC in the family, and advertise other ongoing programs for siblings and families.	An opportunity for parents and siblings (either in separate groups or together) to ask questions, including to a panel of adult siblings about their experiences. Workshops were provided both online and virtually.
Films and Videos	One hospital in Ontario.	7 (23–29)	Public	To share about the experiences of families, including the perspectives of siblings and parents, about growing up with a child with a CHC.	The types of CHCs covered in the films and videos included autism spectrum disorder, Down syndrome, brain damage, and Type 1 diabetes.
News Articles					
Announcements	One hospital in Ontario.	2 (30,31)	Public	To share information about results, which included a donation to create a centre in Canada to support adults with disabilities, a partnership to build rehabilitation capacity for children with a CHC between institutions, and the winner of a “filmpossible” award.	Donation and partnership were inspired by the experience of a family with an individual with a CHC. Stories about the experiences of siblings of an individual with a CHC shared in the announcement.
Awareness about CHCs	Two hospitals in Ontario and	6 (32–37)	Public	To raise awareness on specific days to appreciate different disabilities and	Each document described the awareness of different days: • Sibling Appreciation Day

	one hospital in Quebec.			roles, in which siblings have a part in the awareness of CHCs. s	• Purple Day for Epilepsy • Cerebral Palsy Awareness Month • Childhood Cancer Awareness Month • Children's Grief Awareness Day • International Day of Persons with Disabilities To raise awareness of these different days, siblings shared stories about their brother or sister with a CHC.
Events	One hospital in Ontario and one hospital in Quebec.	5 (37–41)	Public	To raise awareness about the stories and roles of siblings.	Events included advocacy for individuals with disabilities, as well as events for siblings to attend.
Research Studies	Two hospitals in Ontario and one hospital in Quebec.	10** (42–49)	Public.	To share the findings of research studies.	The topics of research studies included genetic information for siblings of individuals with ASD, genetic mutation that lead to lymphoma, survey findings about the experiences of siblings of a brother or sister with a developmental disability, successful donor liver transplant between siblings, rare autoinflammatory disease based on research of two siblings with juvenile idiopathic arthritis, immunotherapy, trials about accessible equipment, and accessible video games for children with disabilities.
Participation in Research	One hospital in Ontario and one health centre in Nova Scotia.	4* (50,51)	Public	To recruit participants for a research study.	The topics of the research studies were assessments of early behavioural signs of autism spectrum disorder in infants and the experiences of sibling including their needs and feelings.
Stories	Health service provider in British Columbia, three	7 (52–58)	Public	To share the stories of families who have an individual with a CHC.	Stories were shared by mothers and siblings about their experiences with a person with a CHC. For some sibling relationships, both siblings had a CHC, and shared how they supported each other.

	hospitals in Ontario.				
Interviews					
With Siblings	Three hospitals in Ontario and one hospital in Quebec.	27***	Public	To share about the experiences of siblings of a child with a CHC.	Key topics discussed during the interviews and blogs are presented in Table 2.
With Parents	One hospital in Ontario.	12***	Public	To share about the parents' experiences when they have multiple children including a child with a CHC.	
Blogs					
Written by Siblings	Three hospitals in Ontario.	18***	Public	To share key messages from siblings about their experiences when they have a sibling with a CHC.	
Written by Parents	Two hospitals in Ontario and a health service provider in British Columbia.	13***	Public	To share the stories of families of a child with a CHC and the siblings from the parents' perspective.	

*Programs, workshops, and participation in research studies are advertised and updated on an ongoing basis on the websites of the children's hospital and/or treatment centres. The number of documents refers to the advertisements and newsletters that was posted on the websites.

**Two documents are no longer available on the website, but was included in the analysis.

*** References were not provided in order to ensure confidentiality of the authors and families mentioned in the blogs and interviews.

References for Table 1

1. Holland Bloorview Kids Rehabilitation Hospital. SibKit [Internet]. [cited 2021 Jun 2]. Available from: https://hollandbloorview.ca/sites/default/files/2020-06/ABI-SibKit-May2020-Accessible.pdf
2. Holland Bloorview Kids Rehabilitation Hospital. SibKits [Internet]. [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/sibkits
3. Kinross L. A child's brain injury has ripple effects on siblings. SibKit is a safe space to explore them [Internet]. 2019 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/childs-brain-injury-has-ripple-effects-siblings-sibkit-safe-space
4. Holland Bloorview Kids Rehabilitation Hospital. Parent tipsheet: Supporting siblings [Internet]. 2018 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/parent-0
5. BC Children's Hospital. Tips to help your child cope with a brother's or sister's surgery [Internet]. [cited 2021 May 29]. Available from: http://www.bcchildrens.ca/Child-Life-site/Documents/Surgery tip sheet- SIBLINGS.docx
6. Holland Bloorview Kids Rehabilitation Hospital. Tips for inpatient siblings [Internet]. [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/tips
7. Timmons V, Breitenbach M, MacIsaac M. A Resource Guide for Parents of Children with Autism: Supporting Inclusive Practice [Internet]. 2016 [cited 2021 Jun 2]. Available from: https://www.princeedwardisland.ca/sites/default/files/publications/eelc_autism_guide_for_parents.pdf
8. Timmons V, Breitenbach M, MacIsaac M. Educating Children about Autism in an Inclusive Classroom [Internet]. 2016 [cited 2021 Jun 2]. Available from: https://www.princeedwardisland.ca/en/publication/educating-children-about-autism-inclusive-classroom
9. Children's Hospital - London Health Sciences Centre. Helping Schools Cope With Childhood Cancer [Internet]. 2011 [cited 2021 Jun 2]. Available from: https://www.lhsc.on.ca/media/1491/download
10. Kinross L. A twin's bond sparks brilliance [Internet]. 2015 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/twins-bond-sparks-brilliance
11. Kinross L. New book explores sibling emotions about disability [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://www.hollandbloorview.ca/stories-news-events/BLOOM-Blog/new-book-explores-sibling-emotions-about-disability
12. Holland Bloorview Kids Rehabilitation Hospital. "Why do you love Jacob more than us?" | Holland Bloorview [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/why-do-you-love-jacob-more-us
13. Holland Bloorview Kids Rehabilitation Hospital. Around the world, brothers and sisters are no longer an afterthought in a disabled child's care [Internet]. 2019 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/around-world-brothers-and-sisters-are-no-longer-afterthought

14. Holland Bloorview Kids Rehabilitation Hospital. Everything you want to know about siblings [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/everything-you-want-know-about-siblings
15. Children's Hospital of Eastern Ontario. Hospitalization and surgery [Internet]. [cited 2021 Mar 14]. Available from: https://www.cheo.on.ca/en/resources-and-support/hospitalization-and-surgery.aspx#Books
16. Kinross L. BBC Ouch: Brothers, sisters and disability [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://www.hollandbloorview.ca/stories-news-events/BLOOM-Blog/bbc-ouch-brothers-sisters-and-disability
17. Children's Treatment Network. Events [Internet]. [cited 2021 May 30]. Available from: https://www.ctnsy.ca/Events.aspx
18. Holland Bloorview Kids Rehabilitation Hospital. Family workshops and resources [Internet]. [cited 2021 May 30]. Available from: https://hollandbloorview.ca/services/family-workshops-resources
19. Children's Hospital - London Health Sciences Centre. Hematology and oncology [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.lhsc.on.ca/hematology-oncology
20. McMaster Children's Hospital. Autism program [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.hamiltonhealthsciences.ca/mcmaster-childrens-hospital/areas-of-care/developmental-pediatrics-and-rehabilitation/autism-program/
21. Bayshore HealthCare. Canada's young caregivers [Internet]. 2019 [cited 2021 May 30]. Available from: https://www.bayshore.ca/2019/07/19/canadas-young-caregivers/#:~:text=In Toronto%2C The Young Carers,illness%2C or language barrier."
22. SickKids. Women's auxiliary volunteers (WAV) [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.sickkids.ca/en/careers-volunteer/volunteering/womens-auxiliary-volunteers/#bravery
23. Kinross L. A brother with Down syndrome is the focus of the film "Music and Clowns" [Internet]. 2020 [cited 2021 May 29]. Available from: https://research.hollandbloorview.ca/stories-news-events/BLOOM-Blog/brother-down-syndrome-focus-film-music-and-clowns
24. Holland Bloorview Kids Rehabilitation Hospital. A magical bond [Internet]. 2020 [cited 2021 May 29]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2014/03/a-magical-bond_12.html
25. Kinross L. These parents took a year off to learn to "speak Oskar" [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/these-parents-took-year-learn-speak-oskar
26. Kinross L. When a child has a chronic illness, every family member adapts [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/when-child-has-chronic-illness-every-family-member-adapts
27. Holland Bloorview Kids Rehabilitation Hospital. "The people that have the voice are too tired to raise their voice" [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/people-have-voice-are-too-tired-raise-their-voice
28. BBC. My autism and me [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/my-autism-and-me

29. Kinross L. Single mom embraces “life of triage” with autistic boys [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/single-mom-embraces-life-triage-autistic-boys
30. Kinross L. New hub to address developmental disabilities, mental illness [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/new-hub-address-developmental-disabilities-mental-illness
31. Holland Bloorview Kids Rehabilitation Hospital. Director Owen McIntyre’s film “My Life With My Brother Rhys” wins the filmpossible award, presented by Holland Bloorview in partnership TIFF Kids International Film Festival [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/news/director-owen-mcintyres-film-my-life-my-brother-rhys-wins-filmpossible
32. Holland Bloorview Kids Rehabilitation Hospital. Malakhai and Nathaniel [Internet]. [cited 2021 Mar 12]. Available from: https://foundation.hollandbloorview.ca/stories-news-events/stories/malakhai-and-nathaniel
33. Montreal Children’s Hospital. Seizing the moment: Sarah’s journey [Internet]. [cited 2021 Mar 12]. Available from: https://www.thechildren.com/patients-families/patient-testimonials/seizing-moment-sarahs-journey
34. CBC News. Hospital for Sick Children’s celebrates patients’ siblings [Internet]. 2015 [cited 2021 Jun 2]. Available from: https://www.cbc.ca/news/canada/toronto/hospital-for-sick-children-celebrates-patients-siblings-1.3103361
35. SickKids. “Grieving in chunks”: helping kids cope with the death of a loved one. 2017.
36. Holland Bloorview Kids Rehabilitation Hospital. After the fall [Internet]. 2014 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/after-fall
37. SickKids. Photovoice: The power of photography brings teen cancer patients and siblings together [Internet]. 2019 [cited 2021 Mar 12]. Available from: https://www.sickkids.ca/en/news/archive/2019/photovoice-the-power-of-photography-brings-teen-cancer-patients-and-siblings-together-/
38. Holland Bloorview Kids Rehabilitation Hospital. A voice for Andrew [Internet]. 2013 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/voice-andrew
39. Montreal Children’s Hospital. Former volunteer brings siblings together at the Just for Kids Sibling Park [Internet]. 2019 [cited 2021 May 30]. Available from: https://www.thechildren.com/news-and-events/latest-news/former-volunteer-brings-siblings-together-just-kids-sibling-park
40. Kinross L. Young carers and other pieces of disability news [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/young-carers-and-other-pieces-disability-news
41. Montreal Children’s Hospital. Sibling drop-off program launches in the Neonatal Intensive Care Unit (NICU) [Internet]. 2017 [cited 2021 May 30]. Available from: https://www.thechildren.com/news-and-events/latest-news/sibling-drop-program-launches-neonatal-intensive-care-unit-nicu
42. SickKids. Genome testing for siblings of individuals with autism may detect diagnosis before symptoms appear [Internet]. 2019 [cited 2021 Jun 2]. Available from: https://www.sickkids.ca/en/news/archive/2019/genome-testing-for-siblings-of-individuals-with-autism-may-detect-diagnosis-before-symptoms-appear/

43. Montreal Children’s Hospital. Seeing double: siblings diagnosed with same rare disease [Internet]. 2018 [cited 2021 May 30]. Available from: https://www.thechildren.com/news-and-events/latest-news/seeing-double-siblings-diagnosed-same-rare-disease
44. Kinross L. Siblings say depression in disabled adults is their top worry [Internet]. 2018 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/siblings-say-depression-disabled-adults-their-top-worry
45. SickKids. SickKids-led team sheds more light on new autoinflammatory disease [Internet]. 2018 [cited 2021 May 30]. Available from: https://www.sickkids.ca/en/news/archive/2018/sickkids-led-team-sheds-more-light-on-new-autoinflammatory-disease-/
46. Holland Bloorview Kids Rehabilitation Hospital. Here’s one video game parents will welcome into their home [Internet]. [cited 2021 May 30]. Available from: https://foundation.hollandbloorview.ca/stories-news-events/stories/heres-one-video-game-parents-will-welcome-their-home
47. Jones M. Maritza’s dream: Games that make therapy fun [Internet]. 2015 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/maritzas-dream-games-make-therapy-fun
48. Montreal Children’s Hospital. Scientific breakthrough: promising new target for immunotherapy [Internet]. 2018 [cited 2021 May 30]. Available from: https://muhc.ca/newsroom/news/scientific-breakthrough-promising-new-target-immunotherapy
49. Sharp MK. Taking steps, together, with “Upsee” [Internet]. 2014 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2014/04/taking-steps-together-with-upsee.html
50. IWK Health Centre. Active research areas [Internet]. [cited 2021 May 30]. Available from: https://www.iwk.nshealth.ca/research/active-research-areas
51. Holland Bloorview Kids Rehabilitation Hospital. Participate in research [Internet]. [cited 2021 May 30]. Available from: https://research.hollandbloorview.ca/get-involved-give/participate-in-research
52. Kinross L. How difficult could caregiving be? This sister found out [Internet]. 2016 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2016/02/how-difficult-could-caregiving-be-this.html
53. Holland Bloorview Kids Rehabilitation Hospital. Isn’t a person more than a brain? [Internet]. 2013 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2013/07/isnt-person-more-than-brain.html
54. Montreal Children’s Hospital. One breath at a time [Internet]. [cited 2021 May 30]. Available from: https://www.thechildren.com/patients-families/patient-testimonials/one-breath-time
55. Hamilton Health Sciences. Opting for an ostomy: a young woman’s journey with Crohn’s [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://www.hamiltonhealthsciences.ca/share/abby-colling/
56. Provincial Health Services Authority. Rosalind’s story [Internet]. [cited 2021 May 30]. Available from: http://www.phsa.ca/health-info/hearing-loss-early-language/stories-from-families/rosalinds-story
57. Holland Bloorview Kids Rehabilitation Hospital. Special needs parents: Here’s how to

make sure your other kids are supported [Internet]. 2020 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/news/special-needs-parents-heres-how-make-sure-your-other-kids-are-supported

58. Reid K. The inherited metabolic disorders news [Internet]. 2011 [cited 2021 Jun 2]. Available from: https://www.lhsc.on.ca/media/2486/download

References for Full Manuscript

1. Perrin JM, Bloom SR, Gortmaker SL. The increase of childhood chronic conditions in the United States. JAMA. 2007;297(24):2755.
2. Newacheck PW, Strickland B, Shonkoff JP, Perrin JM, McPherson M, McManus M, et al. An epidemiologic profile of children with special health care needs. Pediatrics. 1998;102(1):117–23.
3. Knecht C, Hellmers C, Metzing S. The perspective of siblings of children with chronic illness: a literature review. J Pediatr Nurs. 2015 Jan 1;30(1):102–16.
4. Stein REK, Jessop DJ. A noncategorical approach to chronic childhood illness. Public Health Rep. 1982;97(4):354–62.
5. King S, Teplicky R, King G, Rosenbaum P. Family-centered service for children with cerebral palsy and their families: A review of the literature. Semin Pediatr Neurol. 2004;11(1):78–86.
6. Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. Am Psychol. 2000;55(5):469–80.
7. Stewart DA, Law MC, Rosenbaum P, Willms DG, Stewart DA, Law MC, et al. A qualitative study of the transition to adulthood for youth with physical disabilities. Phys Occup Ther Pediatr. 2009;21(4):3–21.
8. Gorter JW, Stewart D, Woodbury-Smith M. Youth in transition: care, health and development. Child Care Health Dev. 2011 Nov 1;37(6):757–63.
9. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Can J Community Ment Heal. 2014;33(1):45–61.
10. Rosenbaum P, King S, Law M, King G, Evans J. Family-centred service. Phys Occup Ther Pediatr. 1998;18(1):1–20.
11. Atkin K, Tozer R. Personalisation, family relationships and autism: Conceptualising the role of adult siblings. J Soc Work. 2014;14(3):225–42.
12. Moyson T, Roeyers H. "The overall quality of my life as a sibling is all right, but of course, it could always be better". Quality of life of siblings of children with intellectual disability: the siblings' perspectives. J Intellect Disabil Res. 2012;56(1):87–101.
13. Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. Intellect Dev Disabil. 2009;47(3):208–19.
14. Bowen M. Clinical view of the family. Fam Syst. 1995;2:153–6.
15. Whiteman SD, McHale SM, Soli A. Theoretical perspectives on sibling relationships. J Fam Theory Rev. 2011;3(2):124–39.
16. Corsano P, Musetti A, Guidotti L, Capelli F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. J Intellect Dev Disabil. 2017;42(2):151–61.
17. Latta A, Rampton T, Rosemann J, Peterson M, Mandleco B, Dyches T, et al. Snapshots reflecting the lives of siblings of children with autism spectrum disorders. Child Care Health Dev. 2014;40(4):515–24.
18. McHale SM, Sloan J, Simeonsson RJ. Sibling relationships or children with autistic, mentally retarded, and nonhandicapped brothers and sisters. J Autism Dev Disord. 1986;16(4):399–413.
19. McHale SM, Updegraff KA, Feinberg ME. Siblings of youth with autism spectrum disorders: theoretical perspectives on sibling relationships and individual adjustment. J

Autism Dev Disord. 2016;46(2):589–602.

20. Davys D, Haigh C. Older parents of people who have a learning disability: perceptions of future accommodation needs. Br J Learn Disabil. 2008;36(1):66–72.
21. The Sibling Collaborative. Understanding the sibling experience: sibling needs assessment survey 2017 [Internet]. 2018 [cited 2021 Jun 2]. p. 1–19. Available from: http://cdn.agilitycms.com/partners-for-planning/Understanding the Sibling Experience - March 2018 - Final.pdf
22. Redquest BK, Tint A, Ries H, Goll E, Rossi B, Lunsky Y. Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. J Policy Pract Intellect Disabil. 2020;17(3):239–46.
23. Mascha K, Boucher J. Preliminary investigation of a qualitative method of examining siblings' experiences of living with a child with ASD. Br J Dev Disabil. 2006;52(1):19–28.
24. Arnold CK, Heller T, Kramer J. Support needs of siblings of people with developmental disabilities. Intellect Dev Disabil. 2012;50(5):373–82.
25. Krauss MW, Seltzer MM, Gordon R, Friedman DH. Binding ties: the roles of adult siblings of persons with mental retardation. Ment Retard. 1996;34(2):83–93.
26. Sibling Support Project. The Sibling Support Project [Internet]. 2020 [cited 2020 Nov 15]. Available from: https://www.siblingsupport.org/
27. Johnson AB. Sibshops: A follow-up of participants of a sibling support program. University of Washington; 2005.
28. D'Arcy F, Flynn J, McCarthy Y, O'Connor C, Tierney E. Sibshops. An evaluation of an interagency model. J Intellect Disabil. 2005;9(1):43–57.
29. Opening Hearts. SibTeens [Internet]. 2019 [cited 2021 Jun 2]. Available from: https://www.openinghearts.ca/sibteens/
30. Cutrona CE. Social support principles for strengthening families: messages from the USA. In: Canavan J, Dolan P, Pinkerton J, editors. Family Support: Direction from Diversity. London: Jessica Kingsley; 2000. p. 103–22.
31. Arnold CK, Heller T. Caregiving experiences and outcomes: Wellness of adult siblings of people with intellectual disabilities. Curr Dev Disord Reports. 2018;5(3):143–9.
32. Dyke P, Mulroy S, Leonard H. Siblings of children with disabilities: challenges and opportunities. Acta Paediatr Int J Paediatr. 2009;98(1):23–4.
33. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. J Appl Res Intellect Disabil. 2018;31(3):423–34.
34. Keim-Malpass J, Baernholdt M, Erickson JM, Ropka ME, Schroen AT, Steeves RH. Blogging through cancer: Young women's persistent problems shared online. Cancer Nurs. 2013 Mar;36(2):163–72.
35. Marcus MA, Westra HA, Eastwood JD, Barnes KL, Group MMR. What are young adults saying about mental health? an analysis of internet blogs. J Med Internet Res. 2012 Jan 30;14(1):e1868.
36. Clarke JN, Lang L. Mothers whose children have ADD/ADHD discuss their children's medication use: An investigation of blogs. Soc Work Health Care. 2012 May;51(5):402–16.
37. Rice RE. Influences, usage, and outcomes of Internet health information searching: multivariate results from the Pew surveys. Int J Med Inform. 2006;75(1):8–28.

38. Alsem MW, Ausems F, Verhoef M, Jongmans MJ, Meily-Visser JMA, Ketelaar M. Information seeking by parents of children with physical disabilities: an exploratory qualitative study. Res Dev Disabil. 2017;60:125–34.
39. Siblings Australia [Internet]. [cited 2021 Jul 22]. Available from: https://siblingsaustralia.org.au/
40. Sibs [Internet]. [cited 2021 Jul 22]. Available from: https://www.sibs.org.uk/
41. Sibling Leadership Network [Internet]. [cited 2021 Jul 22]. Available from: https://siblingleadership.org/
42. The Sibling Collaborative [Internet]. [cited 2021 Jul 22]. Available from: https://sibcollab.ca/
43. Ontario Brain Institute. Ways community members can participate in the stages of research. 2019.
44. Smits D-W, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. Res Involv Engagem. 2020;6(1):30.
45. Bowen GA. Document analysis as a qualitative research method. Qual Res J. 2009;9(2):27–40.
46. Children's Healthcare Canada. Our members and patrons [Internet]. 2021 [cited 2021 Apr 7]. Available from: https://www.childrenshealthcarecanada.ca/members-patrons
47. Orr E, Jack S, Sword W, Ireland S, Ostolosky L. Understanding the blogging practices of women undergoing in vitro fertilization (IVF): a discourse analysis of women's IVF blogs. Qual Rep. 2017;22(8).
48. Altheide DL, Schneider CJ. Process of document analysis. In: Qualitative Media Analysis. 2nd ed. Thousand Oaks, California: Sage Publications Inc.; 2013. p. 39–74.
49. Hsieh H-F, Shannon SE. Three approaches to qualitative content analysis. Qual Health Res. 2005 Nov 1;15(9):1277–88.
50. Bowen GA. Supporting a grounded theory with an audit trail: an illustration. Int J Soc Res Methodol. 2009 Oct;12(4):305–16.
51. Lincoln Y, Guba EG. Naturalistic inquiry. Beverly Hills: SAGE; 1985.
52. Krefting L. Rigor in qualitative research: The assessment of trustworthiness. Am J Occup Ther. 1991 Mar 1;45(3):214–22.
53. Moser A, Korstjens I. Series: Practical guidance to qualitative research. Part 3: Sampling, data collection and analysis. Eur J Gen Pract. 2018 Jan 4;24(1):9–18.
54. Patton MQ. Enhancing the quality and credibility of qualitative analysis. Health Serv Res. 1999 Dec;34(5 Pt 2):1189–208.
55. Kinross L. A twin's bond sparks brilliance [Internet]. 2015 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/twins-bond-sparks-brilliance
56. Kinross L. New book explores sibling emotions about disability [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://www.hollandbloorview.ca/stories-news-events/BLOOM-Blog/new-book-explores-sibling-emotions-about-disability
57. Holland Bloorview Kids Rehabilitation Hospital. "Why do you love Jacob more than us?" | Holland Bloorview [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/why-do-you-love-jacob-more-us
58. Holland Bloorview Kids Rehabilitation Hospital. Around the world, brothers and sisters

are no longer an afterthought in a disabled child's care [Internet]. 2019 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/around-world-brothers-and-sisters-are-no-longer-afterthought
59. Holland Bloorview Kids Rehabilitation Hospital. Everything you want to know about siblings [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/everything-you-want-know-about-siblings
60. Children's Hospital of Eastern Ontario. Hospitalization and surgery [Internet]. [cited 2021 Mar 14]. Available from: https://www.cheo.on.ca/en/resources-and-support/hospitalization-and-surgery.aspx#Books
61. Kinross L. BBC Ouch: Brothers, sisters and disability [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://www.hollandbloorview.ca/stories-news-events/BLOOM-Blog/bbc-ouch-brothers-sisters-and-disability
62. Children's Treatment Network. Events [Internet]. [cited 2021 May 30]. Available from: https://www.ctnsy.ca/Events.aspx
63. Holland Bloorview Kids Rehabilitation Hospital. Family workshops and resources [Internet]. [cited 2021 May 30]. Available from: https://hollandbloorview.ca/services/family-workshops-resources
64. Children's Hospital - London Health Sciences Centre. Hematology and oncology [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.lhsc.on.ca/hematology-oncology
65. McMaster Children's Hospital. Autism program [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.hamiltonhealthsciences.ca/mcmaster-childrens-hospital/areas-of-care/developmental-pediatrics-and-rehabilitation/autism-program/
66. Bayshore HealthCare. Canada's young caregivers [Internet]. 2019 [cited 2021 May 30]. Available from: https://www.bayshore.ca/2019/07/19/canadas-young-caregivers/#:~:text=In Toronto%2C The Young Carers,illness%2C or language barrier."
67. SickKids. Women's auxiliary volunteers (WAV) [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.sickkids.ca/en/careers-volunteer/volunteering/womens-auxiliary-volunteers/#bravery
68. Kinross L. A brother with Down syndrome is the focus of the film "Music and Clowns" [Internet]. 2020 [cited 2021 May 29]. Available from: https://research.hollandbloorview.ca/stories-news-events/BLOOM-Blog/brother-down-syndrome-focus-film-music-and-clowns
69. Holland Bloorview Kids Rehabilitation Hospital. A magical bond [Internet]. 2020 [cited 2021 May 29]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2014/03/a-magical-bond_12.html
70. Kinross L. These parents took a year off to learn to "speak Oskar" [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/these-parents-took-year-learn-speak-oskar
71. Kinross L. When a child has a chronic illness, every family member adapts [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/when-child-has-chronic-illness-every-family-member-adapts
72. Holland Bloorview Kids Rehabilitation Hospital. "The people that have the voice are too tired to raise their voice" [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/people-have-voice-are-

too-tired-raise-their-voice

73. BBC. My autism and me [Internet]. 2014 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/my-autism-and-me
74. Kinross L. Single mom embraces “life of triage” with autistic boys [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/single-mom-embraces-life-triage-autistic-boys
75. Kinross L. New hub to address developmental disabilities, mental illness [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/new-hub-address-developmental-disabilities-mental-illness
76. Holland Bloorview Kids Rehabilitation Hospital. Director Owen McIntyre’s film “My Life With My Brother Rhys” wins the filmpossible award, presented by Holland Bloorview in partnership TIFF Kids International Film Festival [Internet]. 2016 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/news/director-owen-mcintyres-film-my-life-my-brother-rhys-wins-filmpossible
77. Holland Bloorview Kids Rehabilitation Hospital. Malakhai and Nathaniel [Internet]. [cited 2021 Mar 12]. Available from: https://foundation.hollandbloorview.ca/stories-news-events/stories/malakhai-and-nathaniel
78. Montreal Children’s Hospital. Seizing the moment: Sarah’s journey [Internet]. [cited 2021 Mar 12]. Available from: https://www.thechildren.com/patients-families/patient-testimonials/seizing-moment-sarahs-journey
79. CBC News. Hospital for Sick Children’s celebrates patients’ siblings [Internet]. 2015 [cited 2021 Jun 2]. Available from: https://www.cbc.ca/news/canada/toronto/hospital-for-sick-children-celebrates-patients-siblings-1.3103361
80. SickKids. “Grieving in chunks”: helping kids cope with the death of a loved one. 2017.
81. Holland Bloorview Kids Rehabilitation Hospital. After the fall [Internet]. 2014 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/after-fall
82. SickKids. Photovoice: The power of photography brings teen cancer patients and siblings together [Internet]. 2019 [cited 2021 Mar 12]. Available from: https://www.sickkids.ca/en/news/archive/2019/photovoice-the-power-of-photography-brings-teen-cancer-patients-and-siblings-together-/
83. Holland Bloorview Kids Rehabilitation Hospital. A voice for Andrew [Internet]. 2013 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/voice-andrew
84. Montreal Children’s Hospital. Former volunteer brings siblings together at the Just for Kids Sibling Park [Internet]. 2019 [cited 2021 May 30]. Available from: https://www.thechildren.com/news-and-events/latest-news/former-volunteer-brings-siblings-together-just-kids-sibling-park
85. Kinross L. Young carers and other pieces of disability news [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/young-carers-and-other-pieces-disability-news
86. Montreal Children’s Hospital. Sibling drop-off program launches in the Neonatal Intensive Care Unit (NICU) [Internet]. 2017 [cited 2021 May 30]. Available from: https://www.thechildren.com/news-and-events/latest-news/sibling-drop-program-launches-neonatal-intensive-care-unit-nicu
87. SickKids. Genome testing for siblings of individuals with autism may detect diagnosis

before symptoms appear [Internet]. 2019 [cited 2021 Jun 2]. Available from: https://www.sickkids.ca/en/news/archive/2019/genome-testing-for-siblings-of-individuals-with-autism-may-detect-diagnosis-before-symptoms-appear/
88. Montreal Children's Hospital. Seeing double: siblings diagnosed with same rare disease [Internet]. 2018 [cited 2021 Mar 12]. Available from: https://www.thechildren.com/news-and-events/latest-news/seeing-double-siblings-diagnosed-same-rare-disease
89. Kinross L. Siblings say depression in disabled adults is their top worry [Internet]. 2018 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/siblings-say-depression-disabled-adults-their-top-worry
90. SickKids. SickKids-led team sheds more light on new autoinflammatory disease [Internet]. 2018 [cited 2021 May 30]. Available from: https://www.sickkids.ca/en/news/archive/2018/sickkids-led-team-sheds-more-light-on-new-autoinflammatory-disease-/
91. Holland Bloorview Kids Rehabilitation Hospital. Here's one video game parents will welcome into their home [Internet]. [cited 2021 May 30]. Available from: https://foundation.hollandbloorview.ca/stories-news-events/stories/heres-one-video-game-parents-will-welcome-their-home
92. Jones M. Maritza's dream: Games that make therapy fun [Internet]. 2015 [cited 2021 Mar 12]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/maritzas-dream-games-make-therapy-fun
93. Montreal Children's Hospital. Scientific breakthrough: promising new target for immunotherapy [Internet]. 2018 [cited 2021 May 30]. Available from: https://muhc.ca/newsroom/news/scientific-breakthrough-promising-new-target-immunotherapy
94. Sharp MK. Taking steps, together, with "Upsee" [Internet]. 2014 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2014/04/taking-steps-together-with-upsee.html
95. IWK Health Centre. Active research areas [Internet]. [cited 2021 May 30]. Available from: https://www.iwk.nshealth.ca/research/active-research-areas
96. Holland Bloorview Kids Rehabilitation Hospital. Participate in research [Internet]. [cited 2021 May 30]. Available from: https://research.hollandbloorview.ca/get-involved-give/participate-in-research
97. Kinross L. How difficult could caregiving be? This sister found out [Internet]. 2016 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2016/02/how-difficult-could-caregiving-be-this.html
98. Holland Bloorview Kids Rehabilitation Hospital. Isn't a person more than a brain? [Internet]. 2013 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2013/07/isnt-person-more-than-brain.html
99. Montreal Children's Hospital. One breath at a time [Internet]. [cited 2021 May 30]. Available from: https://www.thechildren.com/patients-families/patient-testimonials/one-breath-time
100. Hamilton Health Sciences. Opting for an ostomy: a young woman's journey with Crohn's [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://www.hamiltonhealthsciences.ca/share/abby-colling/
101. Provincial Health Services Authority. Rosalind's story [Internet]. [cited 2021 May 30].

Available from: http://www.phsa.ca/health-info/hearing-loss-early-language/stories-from-families/rosalinds-story

102. Holland Bloorview Kids Rehabilitation Hospital. Special needs parents: Here's how to make sure your other kids are supported [Internet]. 2020 [cited 2021 May 30]. Available from: https://hollandbloorview.ca/stories-news-events/news/special-needs-parents-heres-how-make-sure-your-other-kids-are-supported
103. Reid K. The inherited metabolic disorders news [Internet]. 2011 [cited 2021 Jun 2]. Available from: https://www.lhsc.on.ca/media/2486/download
104. Holland Bloorview Kids Rehabilitation Hospital. SibKit [Internet]. [cited 2021 Jun 2]. Available from: https://hollandbloorview.ca/sites/default/files/2020-06/ABI-SibKit-May2020-Accessible.pdf
105. Holland Bloorview Kids Rehabilitation Hospital. SibKits [Internet]. [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/sibkits
106. Kinross L. A child's brain injury has ripple effects on siblings. SibKit is a safe space to explore them [Internet]. 2019 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/stories-news-events/BLOOM-Blog/childs-brain-injury-has-ripple-effects-siblings-sibkit-safe-space
107. Timmons V, Breitenbach M, MacIsaac M. A Resource Guide for Parents of Children with Autism: Supporting Inclusive Practice [Internet]. 2016 [cited 2021 Jun 2]. Available from: https://www.princeedwardisland.ca/sites/default/files/publications/eelc_autism_guide_for_parents.pdf
108. Timmons V, Breitenbach M, MacIsaac M. Educating Children about Autism in an Inclusive Classroom [Internet]. 2016 [cited 2021 Jun 2]. Available from: https://www.princeedwardisland.ca/en/publication/educating-children-about-autism-inclusive-classroom
109. Children's Hospital - London Health Sciences Centre. Helping Schools Cope With Childhood Cancer [Internet]. 2011 [cited 2021 Jun 2]. Available from: https://www.lhsc.on.ca/media/1491/download
110. Holland Bloorview Kids Rehabilitation Hospital. Parent tipsheet: Supporting siblings [Internet]. 2018 [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/parent-0
111. BC Children's Hospital. Tips to help your child cope with a brother's or sister's surgery [Internet]. [cited 2021 May 29]. Available from: http://www.bcchildrens.ca/Child-Life-site/Documents/Surgery tip sheet- SIBLINGS.docx
112. Holland Bloorview Kids Rehabilitation Hospital. Tips for inpatient siblings [Internet]. [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/tips
113. Zhao Y, Zhang J. Consumer health information seeking in social media: a literature review. Health Info Libr J. 2017;34(4):268–83.
114. Freeman M, Stewart D, Cunningham CE, Gorter JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. J Transit Med. 2018;1(1).
115. Dansby RA, Turns B, Whiting JB, Crane J. A phenomenological content analysis of online support seeking by siblings of people with autism. J Fam Psychother.

2018;29(3):181–200.
116. Russell DJ, Sprung J, McCauley D, Kraus de Camargo O, Buchanan F, Gulko R, et al. Knowledge exchange and discovery in the age of social media: the journey from inception to establishment of a parent-led web-based research advisory community for childhood disability. J Med Internet Res. 2016;18(11):e293.
117. Davys D, Mitchell D, Haigh C. Futures planning, parental expectations and sibling concern for people who have a learning disability. J Intellect Disabil. 2010 Sep 9;14(3):167–83.
118. Tozer R, Atkin K. "Recognized, valued and supported"? The experiences of adult siblings of people with autism plus learning disability. J Appl Res Intellect Disabil. 2015;28(4):341–51.
119. Leane M. "I don't care anymore if she wants to cry through the whole conversation, because it needs to be addressed": Adult siblings' experiences of the dynamics of future care planning for brothers and sisters with a developmental disability. J Appl Res Intellect Disabil. 2020;33(5):950–61.
120. Davys D, Mitchell D, Haigh C. Futures planning - adult sibling perspectives. Br J Learn Disabil. 2015;43(3):219–26.
121. Leonard H, Foley K-R, Pikora T, Bourke J, Wong K, McPherson L, et al. Transition to adulthood for young people with intellectual disability: the experiences of their families. Eur Child Adolesc Psychiatry. 2016;25(12):1369–81.
122. Waldboth V, Patch C, Mahrer-Imhof R, Metcalfe A. Living a normal life in an extraordinary way: a systematic review investigating experiences of families of young people's transition into adulthood when affected by a genetic and chronic childhood condition. Int J Nurs Stud. 2016;62:44–59.
123. Goldman B, CBC Radio. Cake, balloons, goodbye: What happens when your special needs child ages out of the system [Internet]. CBC Radio; 2018 [cited 2021 May 9]. Available from: https://www.cbc.ca/radio/whitecoat/cake-balloons-goodbye-what-happens-when-your-special-needs-child-ages-out-of-the-system-1.4528167
124. Joly E. Transition to adulthood for young people with medical complexity: an integrative literature review. J Pediatr Nurs. 2015;30(5):e91–103.
125. Children's Healthcare Canada. Health Hub in Transition [Internet]. 2021 [cited 2021 Mar 31]. Available from: https://www.transitionhub.ca/
126. Cross A, Rosenbaum P, Grahovac D, Brocklehurst J, Kay D, Baptiste S, et al. A web-based knowledge translation resource for families and service providers (the "F-words" in childhood disability knowledge hub): Developmental and pilot evaluation study. JMIR Rehabil Assist Technol. 2018;5(2):e10439.
127. Nguyen T, Henderson D, Stewart D, Hlyva O, Punthakee Z, Gorter JW. You never transition alone! Exploring the experiences of youth with chronic health conditions, parents and healthcare providers on self-management. Child Care Health Dev. 2016;42(4):464–72.

Data Availability Statement

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Supplementary Table 1. Description of resources that provide general information to siblings of individuals with a chronic health condition.

Booklets and books
Books were available for siblings and families, in which some books highlighted the importance of understanding the importance of sibling relationships. At one children's hospital, young siblings can refer to a booklist such as a book told from the perspective of a main character who has questions when their sibling has a CHC and is in the hospital (1). One sibling wrote a book to chronicle her sibling relationship in growing up with her sister with a CHC. In addition, booklets are available to provide guidance to parents and teachers about how to communicate with siblings of someone with a CHC. For example, there was a booklet that described how to create inclusive classrooms and educate children about autism with a list of resources available specific for siblings (2).
Podcast
The stories of sibling relationships are also shared through other forms of media. There was a podcast from the United Kingdom (53) that was recommended, which tells the stories of individuals with a CHC, and one episode shared the stories of siblings in families with a child with a CHC. Personal stories from families, including siblings, are primarily shared through films and videos. Documentary films described the journey of the whole family, and while they only focused on the perspective of parents, there were stories about the relationship between the siblings and child with a CHC.
Programs and workshops
There are advertisements that announced past programs and workshops that were available to families. There were five workshops that were advertised as one-time events, in which one workshop was held for free in-person and organized at a children's hospital in Ontario (105), and four documents from one children's hospital that advertised workshops organized by another organization for siblings who are young carers ages 15-25 years old, with the option for virtual attendance and did not indicate a fee (106). Documents also advertised programs that were available for siblings if they preferred to participate in an extended activity for a few weeks, and advertised on an ongoing basis from four hospitals and two service providers in Ontario [references provided for the websites of ongoing advertisements for these programs, (54–59)]. Among the 34 documents of program advertisements, 16 were in-person, 6 were virtual due to COVID-19, and 12 did not indicate the type of program format. Furthermore, out of the 34 documents, there were 6 documents from two children's hospitals (55,56) that referred to other available programs for siblings. There were 2 sibling support programs that were identified to be free, 2 programs that required a fee, and 9 programs that did not indicate a fee. Support involved not only workshops or programs, but also family events where the whole family was welcome to attend. A children's treatment network hosted these family events at different geographical locations such as preschool playgroups, escape room activity, arts and crafts, and board games, with 6 events that required a nominal fee and 3 events that did not indicate a fee out of a total of 9 advertised events [reference to ongoing advertisements on the website (54)].
News articles
Stories
Stories were published as news articles that were written either by parents (n=1), mothers (n=2) or a sibling (n=3), both a mother and sibling (n=1). Mothers focused on stories of their child's lived experience, parenting multiple children with a CHC or the same CHC, and/or the

roles that other children may assume when there is a child with a CHC. One mother described how her son had the role of being a playmate and supporter, in which her son played therapy games to ensure that her daughter practiced skills from a therapy intervention program (3). The primary topics of the articles authored by siblings included sharing their emotions about their sibling relationship, providing support with healthcare management, and transitioning into different roles as a sibling such as a caregiver. For some siblings, they shared their grief when a sibling with a CHC passed away (4). In some sibling relationships, both siblings had a CHC and they shared how they supported each other with managing each other's healthcare. For example, two siblings had the same CHC and a sister shared her positive experience at a health clinic and encouraged her sister to attend the same clinic (5). Siblings shared their experiences with new roles that they had, such as being a full-time caregiver when the parents were no longer able to. One sister shared that she learned how to be a caregiver by experience, in which she had to find supports, organize finances, and identify funding from the government for her brother with a CHC (6).
Announcements about initiatives
News articles also published announcements about initiatives that were inspired by the stories of siblings. For example, an event was organized by an organization to provide an opportunity for young carers, siblings, along with professionals, policy makers and researchers to have discussions about priorities and actions to support young carers (107). A special event was also held to showcase the products from a program. Young people, both youth with cancer and their siblings participated in a Photovoice program to share their experiences of cancer, and they displayed their photographs during Childhood Cancer Awareness Month at a hospital (108). Six news articles (69–74) shared about different awareness days and months about specific disabilities and health conditions, and siblings were encouraged to share stories about their brother or sister with a CHC on these awareness days and months.
Research studies
The news articles also advertised research studies that were conducted about siblings [n=10, references provided for 8 articles (79,81–83,85,86,109,110), with 2 articles no longer posted on the website, but was included in the data analysis]. The topics of these research studies ranged from genetic studies that were conducted for specific health conditions, such as autism spectrum disorder and lymphoma, to successful organ transplants between siblings, to rehabilitation studies about the effectiveness of assistive equipment. There were also four news articles that advertised about current research studies that were actively recruiting sibling participants from one children's hospital in Ontario (88) and one health center in Nova Scotia (87) at the time of publication, such as early intervention studies as well as a survey to understand the needs and feelings of siblings of youth with a CHC.

References for Supplementary Table 1

1. Children's Hospital - London Health Sciences Centre. Hematology and oncology [Internet]. 2021 [cited 2021 May 30]. Available from: https://www.lhsc.on.ca/hematology-oncology
2. Timmons V, Breitenbach M, MacIsaac M. Educating children about autism in an inclusive classroom [Internet]. 2016 [cited 2021 Jun 2]. Available from: https://www.princeedwardisland.ca/en/publication/educating-children-about-autism-inclusive-classroom
3. Provincial Health Services Authority. Rosalind's story [Internet]. [cited 2021 May 30]. Available from: http://www.phsa.ca/health-info/hearing-loss-early-language/stories-from-families/rosalinds-story
4. Holland Bloorview Kids Rehabilitation Hospital. Isn't a person more than a brain? [Internet]. 2013 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2013/07/isnt-person-more-than-brain.html
5. Hamilton Health Sciences. Opting for an ostomy: a young woman's journey with Crohn's [Internet]. 2017 [cited 2021 Mar 12]. Available from: https://www.hamiltonhealthsciences.ca/share/abby-colling/
6. Kinross L. How difficult could caregiving be? This sister found out [Internet]. 2016 [cited 2021 May 30]. Available from: http://bloom-parentingkidswithdisabilities.blogspot.com/2016/02/how-difficult-could-caregiving-be-this.html

CHAPTER THREE

TITLE: Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: Protocol of a scoping review

AUTHORS: Nguyen L, Bootsma J, Di Rezze B, Jack S, Ketelaar M, & Gorter JW

JOURNAL: BMJ Open

CITATION: Nguyen L, Bootsma J, Di Rezze B, Jack S, Ketelaar M, Gorter JW. Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: Protocol of a scoping review. BMJ Open. 2021;11(7):e053184. doi: 10.1136/bmjopen-2021-053184

NOTE: The manuscript included in this thesis is the final peer-reviewed manuscript. The reuse of this manuscript is allowed pursuant to the terms of the Creative Commons Attribution-NonCommercial 4.0 International (CC BY-NC 4.0) license.

Abstract

Introduction: Siblings share a lifelong bond in their relationship, and they may choose to provide support to their brother or sister with a neurodisability. Previous reviews summarized programs that only focused on the behavioural, emotional and psychological outcomes of the siblings. There is a need to synthesize existing evidence and enhance our understanding about programs for siblings to acquire knowledge, develop skills, and become empowered that can help them to provide support to their brother or sister with a neurodisability. The objective of this review is to identify and map the characteristics and outcomes of programs designed to prepare siblings in their future roles to support their brother or sister with a neurodisability.

Methods and Analysis: This review will be conducted using the Joanna Briggs Institute methodology for scoping reviews. An integrated knowledge translation approach will be used by partnering with the Sibling Youth Advisory Council comprised of siblings of individuals with a disability throughout all review phases. Databases to be searched include PsycINFO, CINAHL, Sociological Abstracts, ERIC, EMBASE, Web of Science, MEDLINE (Ovid), and SPORTDiscus, from date of inception to November 2020. Studies of programs designed for siblings of individuals with neurodisabilities, with no exclusion on the age of siblings or context, and published in English will be included. Extracted data will include details of program structure and content, eligibility criteria and participants, context, study methods, and outcomes. A summary of the results will be presented in a tabular form to provide an overview of the programs with an accompanying narrative summary to address the research questions of this review.

Dissemination: Findings from this review will be shared using dissemination strategies in partnership with the Sibling Youth Advisory Council. We will share the findings with key stakeholders such as healthcare providers, researchers, and patient and family advocacy groups.

Strengths and limitations of this study

- This review will use a systematic approach to synthesize information about programs designed to support siblings in their future roles to individuals with a neurodisability across the lifespan.
- Findings from this review will be limited to siblings of individuals with a neurodisability, and may not be generalizable for other diagnoses.
- Sibling partners will be involved in all phases of the scoping review, to ensure that the findings of this review are relevant and meaningful for stakeholders.

INTRODUCTION

There is an estimated 150 million children and youth under the age of 18 with a disability, including a neurodisability [1]. Neurodisability can be broadly defined as a group of congenital or acquired long-term conditions due to an impairment of the brain and/or neuromuscular system that may create functional limitations [2]. Children and youth with a neurodisability are growing up and becoming adults. During their transition to adulthood, they may take on different opportunities to explore their interests and goals, including school, work, family, and leisure. They might also experience biological, social, and emotional challenges, such as exploring the option to attend postsecondary education, finding employment, developing long-term relationships, and navigating adult healthcare services [3,4]. Even as adults, they may continue to experience challenges with navigating different systems such as healthcare, education, and social services. To navigate these different systems, individuals with neurodisabilities can turn to their families for support during the progression through emerging adulthood [4]. Adults with neurodisabilities will continue to require support, such as with personal care and activities of daily living, as they age [5,6]. Families are often in a position to provide the most optimal support because they have been involved throughout their child's care, in which they know their child best including the child's strengths, areas of improvement and goals [7].

Siblings are an integral part of the family, and every sibling relationship is different with varying levels of emotional closeness, social connectedness, and expectations of each other [8]. Siblings have a unique relationship, in which they share a lifelong bond. When a brother or sister has a neurodisability, a sibling may choose to provide support. Yet, sibling relationships are also highly dynamic and can change over time depending on the needs, roles, and commitments of the whole family [9]. Siblings may choose to support their family in different ways, such as providing emotional support to their brother or sister with a neurodisability or taking on responsibilities to help the whole family [10]. Siblings often become closer when there is planning involved from the whole family with the recognition of the sibling's role for the future during adulthood [11,12]. Sometimes, there is an implicit expectation that the siblings will eventually become carers for their brother or sister with a disability when parents are no longer able to provide support [13]. Some siblings may find that clear and explicit expectations about future responsibilities can be helpful to understand their role [13]. Having a sibling with a neurodisability could affect siblings' own future planning, such as career choice, partner choice, or decision to have children [13]. Many siblings will continue to be a part of the lives of their brother or sister with a neurodisability throughout the lifespan.

Siblings can offer support in different ways to their brother or sister with a neurodisability. There are four main types of support [14]: concrete support that refers to acts of practical assistance; emotional support that includes acts of empathy; advice support that comprises of acts to offer information, emotional reassurance, and guidance; and esteem support that focuses on the reinforcement of personal worth of an individual. Different programs can help to prepare siblings for future roles to support their brother or sister with a neurodisability. These programs can serve different purposes such as sharing information about neurodisabilities, providing opportunities for siblings to connect with each other, and offering resources for the siblings to support their brother or sister with a disability [15,16]. The programs can be tailored for different age groups. For example, Sibshops have been developed in the United States for siblings ages 8 to 13 years old to learn strategies to deal with situations experienced by their brother or sister with a disability [15]. Another program is Sibs Talk, which was developed in the

United Kingdom for students to develop coping strategies, acquire knowledge about their siblings' disability, address challenges about their experiences at home and school, and identify their responsibilities to their siblings with a disability [17]. Family interventions may also include sessions for siblings to learn about the disability of their brother or sister [18] or learn strategies to interact and socialize with their brother or sister that is reinforced by parents [19]. During adulthood, siblings of individuals with a neurodisability might continue to seek supports and resources to address their concerns, such as the mental health of their whole family, housing options and finances [20].

Three systematic reviews have explored interventions and programs for siblings of a brother or sister with a disability [21–23]. Hartling et al. (2010) evaluated the effectiveness of sibling programs that focused on improving behavioural and emotional outcomes in siblings of a brother or sister with a disability and discussed the need for programs to clearly describe the purpose and intended benefits. Tudor and Lerner (2015) provided a review on the nature, content, and outcomes of clinical services designed for siblings of a brother or sister with a developmental disability, and identified how future research can be conducted to identify which siblings might benefit from certain services. McKenzie-Smith (2018) conducted a systematic review to summarize the evidence about the psychological functioning of siblings of a brother or sister with a chronic physical or mental health condition. All three systematic reviews summarized the literature about sibling programs that focused on the behavioural, emotional and psychological outcomes of the siblings themselves. This review will focus on programs designed to prepare siblings in their roles so that they can, in turn, support their brother or sister with a disability. There is a need to synthesize the literature on sibling programs that focus on the knowledge acquisition, skill development, and empowerment of siblings of individuals with a neurodisability.

The aim of this scoping review is to identify and map the characteristics and outcomes for participants in programs designed to prepare siblings in their future roles to support their brother or sister with a neurodisability. A preliminary search on the JBI Database of Systematic Reviews and Implementation Reports, Cochrane Database of Systematic Reviews, PROSPERO, PubMed and CINAHL did not identify any reviews that have summarized the literature about this topic. The results from this review will provide an understanding about existing programs designed for siblings to support their brother or sister with a neurodisability. Results will highlight information about the content of these programs of how siblings can provide support to their brother or sister with a neurodisability. We plan to use key findings from this review to identify current practices, as well as inform the development of resources and tools to support siblings of individuals with neurodisabilities in co-creation with key stakeholders, such as siblings of individuals with neurodisabilities and researchers. Based on recommendations from the UK Medical Research Council, available evidence could be used to inform the development of a resource or intervention [24].

Review questions

Our primary review questions are:

i. What are the characteristics of programs (e.g., purpose, description, eligibility criteria, length, activities, service provider, delivery)?

ii. What are the outcomes for the siblings of individuals with neurodisabilities participating in the programs?

Inclusion criteria

Participants

This review will focus on programs with participants who are siblings of an individual with a neurodisability, defined as a group of congenital or acquired long-term conditions due to an impairment of the brain and/or neuromuscular system that may create functional limitations [2]. Sibling participants may be of varying ages, including children, youth, and adults.

Concept

This review will include studies that describe programs designed to support siblings in their roles. Outcomes of these programs could include, but are not limited to knowledge acquisition, skill development, or empowerment for the siblings. Studies about sibling programs that focus only on therapy or support for the siblings without reference to support the individual with a neurodisability will be excluded. Studies that describe sibling programs without specific objectives targeting siblings of individuals with neurodisabilities will also be excluded.

Context

The context of this review will include all settings that deliver sibling programs, such as school, rehabilitation, healthcare, or community settings, in any country.

Types of sources

This review will consider all study designs including experimental and quasi-experimental study designs including randomized controlled trials, non-randomized controlled trials, before and after studies and interrupted time-series studies. Descriptive studies (e.g., case reports), analytical observational studies (e.g., prospective and retrospective studies, case-control studies and cross-sectional studies) will be included. This review will also consider descriptive observational study designs including case series, individual case reports and descriptive cross-sectional studies for inclusion. Qualitative studies will also be considered that focus on qualitative data including, but not limited to, designs such as phenomenology, grounded theory, ethnography, qualitative description, action research and feminist research. Mixed methods studies will also be considered.

METHODS

This scoping review will be conducted according to the Joanna Briggs Institute (JBI) methodology for scoping reviews [25]. This protocol paper, as well as the final report will be written using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist [26].

Patient and public involvement

This scoping review also uses an integrated knowledge translation as an approach to doing research with knowledge users as equal partners with researchers throughout the study [27]. In this review, we partnered with the Sibling Youth Advisory Council (SibYAC) comprised of six young adult siblings who have a brother or sister with a disability. Based on previous literature, siblings with the lived experiences of having a brother or sister with a disability have often described the importance of being involved in research [28]. The SibYAC identified the relevance of the research questions for this review and program outcomes (e.g., knowledge acquisition, skill development, empowerment) that are important to synthesize in this review. We plan to continue our partnership with the SibYAC throughout the process of conducting this review, such as reviewing preliminary findings and providing recommendations for the interpretation of results, as well as knowledge translation stages such as co-creating reports and presentations or sharing the results with the community.

Search strategy

A three-step search strategy will be employed. An initial limited search of PsycINFO was undertaken on November 17, 2020 to identify articles on the topic. The text words contained in the titles and abstracts of relevant articles, and the index terms used to describe the articles were used to develop a full search strategy for PsycINFO (see Supplementary File 1). The search strategy, including all identified keywords and index terms, will be adapted for each included database and/or information source. The reference list of all included sources of evidence will be screened for additional studies. Studies published in English will be included. Articles published from database inception to the present will be included.

Information sources

The databases to be searched include PsycINFO, Cumulative Index of Nursing and Allied Health Literature (CINAHL), Sociological Abstracts, Education Resources Information Center (ERIC), EMBASE, Web of Science, MEDLINE (Ovid), and Sport Discus.

Study of evidence selection

Following the search, all identified citations will be collated and uploaded into Covidence, systematic review software (Veritas Health Information, Melbourne, Australia) and duplicates will be removed. Following a pilot test, titles and abstracts will then be screened independently by two reviewers (LN and JB) against the inclusion criteria for the review. Potentially relevant sources will be retrieved in full with citation details. The full text of selected studies will be assessed in detail against the inclusion criteria by two independent reviewers (LN and JB). Reasons for exclusion of sources of evidence at full text that do not meet the inclusion criteria will be recorded and reported in the scoping review. Any disagreements that arise between the reviewers at each stage of the selection process will be resolved through discussion or consultation with a third reviewer. The results of the search and the study inclusion process will be reported in full in the final scoping review and presented in a Preferred Reporting Items for Systematic Reviews and Meta-analyses extension for scoping review (PRISMA-ScR) flow diagram [29].

Data extraction

Data extraction from papers included in the scoping review will be performed independently by two reviewers (LN and JB) using a data extraction tool developed by all research team members, including the SibYAC. Data extracted will include the following: author(s), year of publication, country of origin (where the study program was published or conducted), study aim(s), purpose(s) of the program, study population and sample size, methodology/methods, program context/setting, program description, program development (who developed the program, process of developing the program), program delivery (including the provider and type of delivery), duration of the program, program activities, participants' needs and goals while participating in the program, participant outcomes, and program evaluation. A draft of the data extraction sheet is provided in Supplementary File 2. This draft form was created based on the JBI template source of evidence details, characteristics and results extraction instrument (22), as well as the Cochrane Collaboration data extraction template (29). The data extraction sheet will be modified as necessary during the process of extracting data from each included evidence source. Modifications will be detailed in the scoping review. Any disagreements that arise between the reviewers will be resolved through discussion or consultation with a third reviewer. The authors of included articles will be contacted to request missing or additional data based on the data extraction sheet.

Data analysis and presentation

The extracted data will be presented in a tabular form that provides a comprehensive overview about sibling programs based on the information outlined in the data extraction form. There will be an accompanying narrative summary to describe how the results address the research questions of this scoping review.

Overall, siblings can offer support in different ways as they will continue to be a part of their lives to their brother or sister with a neurodisability throughout the lifespan. While siblings may want to offer support, they need to be prepared and empowered to take on this role in the future. This scoping review addresses a gap in the literature about the available evidence about programs designed to support siblings of individuals with a neurodisability in their future roles. The SibYAC will be involved as partners throughout the process of conducting this review to ensure that the findings are relevant and meaningful to the community. Findings from this review will provide a synthesis of evidence-based information about programs as well as identify future directions to inform or enhance existing resources designed to support siblings of individuals with a neurodisability.

DISSEMINATION

The findings from this review will be published in peer-reviewed publications, and presented at local, national, and international conferences. We also plan to share a plain language report with the community. To support our knowledge translation and dissemination activities, we will leverage the infrastructure of a website of a project that has partnered with the SibYAC about the experiences of siblings of youth with a neurodisability [30]. We will post the findings of this review to share with the community. Our research team, including our SibYAC partners, will seek opportunities to share both preliminary and final findings with key stakeholders such as healthcare providers, researchers, patient and family advocacy groups.

ACKNOWLEDGMENTS

This review will contribute towards the doctoral degree for the primary author (LN). We gratefully acknowledge our partnership with the Sibling Youth Advisory Council throughout the process of designing the protocol of this review. The authors would also like to acknowledge the assistance and guidance from Laura Banfield for the development and refinement of the search strategy.

AUTHORS' CONTRIBUTIONS

LN, BDR, SMJ, MK, and JWG contributed to the conceptualization of the review; and LN, JB, BDR, SMJ, MK, and JWG contributed to its design. LN drafted the manuscript, and all authors were involved in the revisions, reviewed the manuscript and provided their final approval.

FUNDING

This review is funded by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream – Phase 1 held by LN. The partnership with the Sibling Youth Advisory Council is financially supported by the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network held by LN. The Scotiabank Chair in Child Health Research is held by JWG.

COMPETING INTERESTS

The authors declare no conflict of interest.

REFERENCES

1 UNICEF. *The state of the world's children 2006: excluded and invisible*. New York: United Nations Children's Fund 2005.

2 Morris C, Janssens A, Tomlinson R, *et al.* Towards a definition of neurodisability: a Delphi survey. *Dev Med Child Neurol* 2013;**55**:1103–8. doi:10.1111/dmcn.12218

3 Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. *Am Psychol* 2000;**55**:469–80. doi:10.1037/0003-066X.55.5.469

4 Freeman M, Stewart D, Cunningham CE, *et al.* Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. *J Transit Med* 2018;**1**. doi:10.1515/jtm-2018-0003

5 Dew A, Llewellyn G, Balandin S. Exploring the later life relationship between adults with cerebral palsy and their non-disabled siblings. *Disabil Rehabil* 2014;**36**:756–64. doi:10.3109/09638288.2013.816786

6 Grossman BR, Magaña S. Introduction to the special issue: Family Support of Persons with Disabilities Across the Life Course. *J Fam Soc Work* 2016;**19**:237–51. doi:10.1080/10522158.2016.1234272

7 Rosenbaum P, King S, Law M, *et al.* Family-centred service. *Phys Occup Ther Pediatr* 1998;**18**:1–20. doi:10.1080/J006v18n01_01

8 Bowen M. Clinical view of the family. *Fam Syst* 1995;**2**:153–6.

9 Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. *Intellect Dev Disabil* 2009;**47**:208–19. doi:10.1352/1934-9556-47.3.208

10 Corsano P, Musetti A, Guidotti L, *et al.* Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *J Intellect Dev Disabil* 2017;**42**:151–61. doi:10.3109/13668250.2016.1226277

11 McHale SM, Sloan J, Simeonsson RJ. Sibling relationships or children with autistic, mentally retarded, and nonhandicapped brothers and sisters. *J Autism Dev Disord* 1986;**16**:399–413. doi:10.1007/BF01531707

12 McHale SM, Updegraff KA, Feinberg ME. Siblings of youth with autism spectrum disorders: theoretical perspectives on sibling relationships and individual adjustment. *J Autism Dev Disord* 2016;**46**:589–602. doi:10.1007/s10803-015-2611-6

13 Davys D, Haigh C. Older parents of people who have a learning disability: perceptions of future accommodation needs. *Br J Learn Disabil* 2008;**36**:66–72. doi:10.1111/j.1468-3156.2007.00447.x

14 Cutrona CE. Social support principles for strengthening families: messages from the USA. In: Canavan J, Dolan P, Pinkerton J, eds. *Family Support: Direction from Diversity*. London: : Jessica Kingsley 2000. 103–22.

15 D'Arcy F, Flynn J, McCarthy Y, *et al.* Sibshops. An evaluation of an interagency model. *J Intellect Disabil* 2005;**9**:43–57. doi:10.1177/1744629505049729

16 Conroy-Harris A. England and Wales. *Adopt Foster* 2019;**43**:91–4. doi:10.1177/0308575919834402

17 Hayden NK, McCaffrey M, Fraser-Lim C, *et al.* Supporting siblings of children with a special educational need or disability: An evaluation of Sibs Talk, a one-to-one intervention delivered by staff in mainstream schools. *Support Learn* 2019;**34**:404–20. doi:10.1111/1467-9604.12275

18 Lobato DJ, Kao BT. Integrated sibling-parent group intervention to improve sibling knowledge and adjustment to chronic illness and disability. *J Pediatr Psychol* 2002;**27**:711–6. doi:10.1093/jpepsy/27.8.711

19 Sheikh R, Patino V, Cengher M, *et al.* Augmenting Sibling Support with Parent-Sibling Training in Families of Children with Autism. *Dev Neurorehabil* 2019;**22**:542–52. doi:10.1080/17518423.2019.1629662

20 Redquest BK, Tint A, Ries H, *et al.* Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. *J Policy Pract Intellect Disabil* 2020;**17**:239–46. doi:10.1111/jppi.12339

21 Hartling L, Milne A, Tjosvold L, *et al.* A systematic review of interventions to support siblings of children with chronic illness or disability. *J Paediatr Child Heal* 2014;**50**:E26-38. doi:10.1111/j.1440-1754.2010.01771.x

22 Tudor ME, Lerner MD. Intervention and support for siblings of youth with developmental disabilities: a systematic review. *Clin Child Fam Psychol Rev* 2015;**18**:1–23. doi:10.1007/s10567-014-0175-1

23 McKenzie Smith M, Pinto Pereira S, Chan L, *et al.* Impact of well-being interventions for siblings of children and young people with a chronic physical or mental health condition: A systematic review and meta-analysis. *Clin Child Fam Psychol Rev* 2018;**21**:246–65. doi:10.1007/s10567-018-0253-x

24 Craig P, Dieppe P, Macintyre S, *et al.* Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ* 2008;**337**:a1655. doi:10.1136/bmj.a1655

25 Peters M, Godfrey C, McInerney P, *et al.* Scoping Reviews. In: Aromataris E, Munn Z, eds. *Joanna Briggs Institute Reviewer's Manual*. JBI Manual for Evidence Synthesis 2020. doi:10.46658/JBIMES-20-12

26 Tricco AC, Lillie E, Zarin W, *et al.* PRISMA extension for scoping reviews (PRISMA-ScR): Checklist and explanation. *Ann Intern Med* 2018;**169**:467–73. doi:10.7326/M18-0850

27 Canadian Institutes of Health Research. Guide to knowledge translation planning at CIHR: Integrated and end-of -grant approaches. 2015.http://www.cihr-irsc.gc.ca/e/45321.html#a3 (accessed 22 Apr 2019).

28 Richardson SLL, Jordan LS. Qualitative inquiry of sibling relationships: reinforcement of disability devaluation through the exclusion of voices. *Disabil Soc* 2017;**32**:1534–54. doi:10.1080/09687599.2017.1351330

29 Moher D, Liberati A, Tetzlaff J, *et al.* Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Med* 2009;**6**:e1000097. doi:10.1371/journal.pmed.1000097

30 CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study. 2021.https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study (accessed 6 May 2021).

Supplementary File 1: Search strategy

PsycINFO (OVID). Search conducted on November 17, 2020.

Example.

Search	Query	Records Retrieved
#1	exp Siblings/ or sibling*.mp.	30852
#2	exp SISTERS/ or sister*.mp.	6420
#3	exp BROTHERS/ or brother*.mp.	6533
#4	disab*.mp.	167345
#5	((neurodevelopmental or intellectual* or developmental*) adj2 (disab* or retard* or impair* or difficult* or disorder*)).mp.	75165
#6	cerebral palsy.mp. or exp Cerebral Palsy/	8350
#7	(cerebral adj palsy).mp.	7984
#8	autism spectrum disorder.mp. or exp Autism Spectrum Disorders/ or Asperger*.mp. or ASD.mp.	48706
#9	pervasive developmental disorder*.mp.	3221
#10	spina bifida.mp. or exp Spina Bifida/	1197
#11	exp EPILEPSY/ or epilepsy.mp.	39992
#12	exp Fetal Alcohol Syndrome/ or fetal alcohol spectrum disorder*.mp. or FASD.mp.	2548
#13	exp Neurodevelopmental Disorders/ or neurodevelopmental disorder.mp.	161860
#14	exp Learning Disabilities/	27715
#15	learning disab*.mp.	27680
#16	exp Stuttering/	4358
#17	stutter*.mp.	6071
#18	exp Attention Deficit Disorder with Hyperactivity/	25804
#19	(attention deficit adj 2 disorder*).mp.	0
#20	ADHD.mp.	29847
#21	developmental coordination disorder*.mp.	1184
#22	developmental co-ordination disorder*.mp.	50
#23	DCD.mp.	946
#24	tic disorder*.mp.	1656
#25	exp Tourette Syndrome/	3201
#26	(tourette* adj2 (disease* or disorder* or syndrome*)).mp.	4230
#27	exp Down's Syndrome/	6325
#28	down* syndrom*.mp.	8427
#29	exp Specific Language Impairment/ or specific language impairment*.mp.	2954
#30	exp Intellectual Development Disorder/	45029
#31	dyscalculia.mp. or exp Acalculia/ or acalculia.mp.	914
#32	(alcohol related adj2 (birth defect* or neurodevelopmental disorder*)).mp.	116
#33	exp Fragile X Syndrome/	1836
#34	Fragile X syndrom*.mp.	2502

#35	fraxa syndrom*.mp.	0
#36	fraxe syndrom*.mp.	0
#37	exp Klinefelters Syndrome/	231
#38	Klinefelter* syndrom*.mp.	385
#39	XXY.mp.	232
#40	Lesch Nyhan syndrom*.mp.	171
#41	Lesch-Nyhan syndrom*.mp.	171
#42	lowe syndrom*.mp.	10
#43	cerebro oculo renal syndrom*.mp.	0
#44	marfan* syndrom*.mp.	79
#45	exp Neurofibromatosis/	16511
#46	neurofibromatos* 1.mp.	356
#47	neurofibromatos* i.mp.	5
#48	Trisomy 21.mp.	421
#49	Noonan syndrom*.mp.	70
#50	exp Prader Willi Syndrome/	546
#51	prader willi syndrom*.mp.	802
#52	exp Rett Syndrome	897
#53	rett* syndrom*.mp.	1323
#54	rett disorder.mp.	27
#55	Rubinstein syndrom*.mp.	0
#56	rubinstein taybi syndrom*.mp.	58
#57	tuberous sclerosis.mp.	700
#58	exp Turners Syndrome/	372
#59	turner* syndrom*.mp.	552
#60	exp Williams Syndrome/	1036
#61	william* syndrom*.mp.	1349
#62	williams-beuren syndrom*.mp.	77
#63	exp Cleft Palate/	491
#64	cleft pal?at*.mp.	819
#65	velo-cardio-facial syndrom*.mp.	156
#66	velocardiofacial syndrom*.mp.	175
#67	exp Trisomy/	211
#68	edward* syndrom*.mp.	7
#69	trisomy 18.mp.	42
#70	exp Dyslexia/	6845
#71	dyslexia*.mp.	10796
#72	(reading adj3 (disabil* or retard* or impair* or difficult* or disorder*)).mp.	12073
#73	clumsy child*.mp.	78
#74	exp Speech Disorders/	29028
#75	speech sound disorder*.mp.	454
#76	exp Articulation Disorders/	1642
#77	articulation disorder*.mp.	1366
#78	phonetic disorder*.mp.	8

#79	exp Language Disorders/	38037
#80	(language adj (disorder* or impair* or defici*)).mp.	13016
#81	disorder language.mp.	47
#82	language development disorder*.mp.	3617
#83	exp Phenylketonuria/	414
#84	phenylketonuria*.mp.	748
#85	PKU.mp.	345
#86	exp Cornelia De Lange Syndrome/	44
#87	cornelia de lange syndrom*.mp.	85
#88	de lange* syndrom*.mp.	105
#89	Duchenne muscular dystrophy.mp.	635
#90	XXY karyotype*.mp.	20
#91	karyotype* xxy.mp.	3
#92	(XXX and (female* or karyotype or chromosome or trisomy)).mp.	86
#93	developmental disabilities/ or learning disorders/ or intellectual disability/ or motor skills disorders.mp.	16064
#94	exp Crying Cat Syndrome/	73
#95	cri du chat syndrom*.mp.	98
#96	happy puppet syndrom*.mp.	3
#97	angelman syndrom*.mp.	436
#98	galactos?emia*.mp.	46
#99	exp Deaf/ or exp Hearing Disorders/ or hearing loss.mp.	23720
#100	exp Blind/ or exp Vision Disorders/	17124
#101	(blind or blindness).mp.	48324
#102	(visual adj1 (impair* or loss or disorder* or disabil* or difficult*)).mp.	5537
#103	program*.mp.	438137
#104	exp Program Development/ or exp Program Evaluation/	28670
#105	resource*.mp.	183369
#106	exp Training/ or train*.mp.	351618
#107	intervention*.mp. or exp Intervention/	413366
#108	educat*.mp.	661227
#109	workshop*.mp.	17726
#110	or/1-3	38256
#111	or/4-102	452686
#112	or/103-109	1470551
#113	and/110-112	1647

Supplementary File 2: Data extraction instrument

<table>
<tr><td colspan="4">Study Information</td></tr>
<tr><td>Date form completed</td><td colspan="3">Identify the date that the form was completed (dd/mm/yyyy).</td></tr>
<tr><td>Name of person extracting data</td><td colspan="3">Identify the reviewer who extracted the data for this form.</td></tr>
<tr><td>Study title</td><td colspan="3">Identify the full title and subtitle.</td></tr>
<tr><td>Authors</td><td colspan="3">Identify all authors as follows: “last name, initial of first name.”</td></tr>
<tr><td>Author affiliations</td><td colspan="3">List affiliations of all authors (e.g., program, department, institution).</td></tr>
<tr><td>Country</td><td colspan="3">Identify the country where the study was conducted.</td></tr>
<tr><td>Study funding source</td><td colspan="3">Describe the funding source for the study and role of funders.</td></tr>
<tr><td>Possible conflicts of interest</td><td colspan="3">Identify possible conflicts of interest declared by authors.</td></tr>
<tr><td colspan="4">Population and Setting</td></tr>
<tr><td>Participant inclusion criteria</td><td colspan="3">Describe the participant inclusion criteria.</td></tr>
<tr><td>Participant exclusion criteria</td><td colspan="3">Describe the participant exclusion criteria.</td></tr>
<tr><td rowspan="5">Participant characteristics</td><td>Total number</td><td>Number of males</td><td>Number of females</td></tr>
<tr><td colspan="3">Age (years)</td></tr>
<tr><td colspan="3">Education level</td></tr>
<tr><td colspan="3">Race/ethnicity</td></tr>
<tr><td colspan="3">Diagnosis of participant’s siblings</td></tr>
<tr><td>Methods of participant recruitment</td><td colspan="3">Identify the different methods of participant recruitment (e.g., social media, word of mouth, participant referral).</td></tr>
<tr><td>Setting context</td><td colspan="3">Describe the environment that the program was implemented (e.g., community, college, university, after school).</td></tr>
<tr><td colspan="4">Methods</td></tr>
<tr><td>Study aim</td><td colspan="3">Identify the study aims.</td></tr>
<tr><td>Type of methodology</td><td colspan="3">Describe the methodology that was used.</td></tr>
<tr><td>Data collection methods</td><td colspan="3">Identify the methods used for data collection (e.g., self-reports, assessment tools, surveys, interviews).</td></tr>
<tr><td>Analysis methods</td><td colspan="3">Identify the methods of analysis (e.g., statistical analyses, qualitative analytic techniques).</td></tr>
<tr><td colspan="4">Program Characteristics</td></tr>
<tr><td>Name</td><td colspan="3">Identify the full name and/or abbreviation.</td></tr>
</table>

Objective(s)	State the objectives of the program.
Duration and frequency	Identify the number and length of sessions, as well as the frequency of sessions.
Mode of delivery	Identify the mode of delivery (e.g., online, in-person).
Start date	Provide the specific start date of the program (month, year).
End date	Provide the specific end date of the program (month, year).
Activities	Identify the activities that were provided in the programs (e.g., weekly meetings, educational workshops, training activities).
Developers	Describe the people who developed the program (e.g., number of developers, background, qualifications, skill level).
Facilitators	Describe the people who facilitated the program (e.g., number of facilitators, background, qualifications, education level).
Participants' needs	Describe the needs of participants in their roles as a sibling while participating in the program.
Participants' goals	Describe the goals of participants in the program and the progress towards achieving these goals.
Outcomes	Report on the outcomes of participants after participating in the program (e.g., satisfaction with the program, knowledge acquisition, skill development, empowerment).
Evaluation	Describe the methods, instruments, and frequency of evaluation assessments.
Sustainability	Describe how the program will be sustained.
Study reference	Provide the full reference for the study.
Other studies of interest	Identify other studies of interest based on the reference list.

CHAPTER FOUR

TITLE: Programs to prepare siblings for future roles to support their sibling with a neurodisability: A scoping review.

AUTHORS: Nguyen L, Bootsma J, Ketelaar M, Di Rezze B, Jack S, & Gorter JW

JOURNAL: Submitted to Current Developmental Disorders Reports.

Abstract

Aims

To identify and map the characteristics and outcomes of programs designed to prepare siblings for their future roles with their sibling with a neurodisability.

Methods

In this scoping review, a systematic search was conducted in eight databases resulting in 5674 non-duplicate articles that were screened. Inclusion criteria were articles that described programs designed to support the roles of typically developing siblings of individuals with a neurodisability. A data extraction form was piloted for 10% of the articles between two independent reviewers, and one reviewer continued to independently extract data from remaining articles. Our research team used a patient-oriented research approach with the Sibling Youth Advisory Council as a partner who provided their perspectives at each review stage.

Results

Fifty-eight articles (published between 1975 to 2020, with >50% published since 2010) met the inclusion criteria, representing 54 sibling programs from 11 countries. Extracted data represented 1033 (553 females) typically developing sibling participants, between 4 to 67 years old. Twenty-seven programs focused on the outcome of knowledge acquisition for the typically developing siblings and thirty-one programs focused on the outcome of empowerment for the typically developing siblings to teach skills to their sibling with a neurodisability.

Conclusions

While there is an increasing number of programs for siblings of individuals with a neurodisability in the past decade, there is a lack of siblings as co-developers or facilitators. Future research should consider the various roles that siblings can have in programs to address their needs.

Keywords: children, youth, siblings, disability, program

INTRODUCTION

Worldwide, there are approximately 150 million children and youth under the age of 18 years with a disability, including a neurodisability [1]. Neurodisability can be broadly defined as a group of congenital or acquired long-term conditions as a result of an impairment of the brain and/or neuromuscular system that may lead to functional limitations [2]. As children and youth with a neurodisability transition to adulthood, they may experience multiple challenges as they navigate developmental trajectories including exploring options for post-secondary education or accessing health services in the adult care system [3,4]. Individuals with neurodisabilities may seek support from their families as they transition into adulthood, including support with personal care and activities of daily living [4-6]. Many families are well-positioned to provide the most optimal support given their history, knowledge, and familiarity of the family member's care and social needs throughout the individual's life [5].

In addition to parental support, in families with more than one child, siblings may also emerge as another source of support for an individual with a neurodisability. Every sibling relationship is unique with differing levels of emotional closeness and expectations of each other [6]. Sibling relationships can evolve or change over time based on the needs, roles, and commitments of the whole family [7]. When a sibling has a neurodisability, the typically developing (TD) sibling may choose to provide support. There are four main types of support: 1) concrete support that includes acts of practical assistance; 2) emotional support that involves acts of empathy; 3) advice support that encompasses acts of provision of information, emotional reassurance, and guidance; and 4) esteem support that includes the reinforcement of the personal worth of an individual [10]. Sometimes, there is an implicit expectation from parents that a TD sibling will be actively involved in supporting the family member with a neurodisability [8].

Given this, TD siblings may need support for different roles that they can assume in supporting their brother or sister [9,10].

Programs are available to support siblings in roles that they may assume. The broad aims of many of these programs include: 1) providing information about neurodisabilities; 2) creating sibling communities to connect and share experiences with each other; or 3) connecting siblings to resources and services to assist them in their supporting role [11,12]. Programs that have been developed have often been tailored to siblings of different ages. Sibshops, for example, was developed in the United States for siblings ages 8 to 13 years old to learn strategies to address situations with their sibling with a disability [11]. Some programs may be targeted for the whole family with specific sessions for siblings to learn about neurodisabilities [13] or to learn strategies to connect with the sibling with a neurodisability. In these programs, parents can be trained on how to reinforce these strategies at home [14].

While descriptions of varied programs exist across the literature, there is limited understanding of the impact and outcomes of these programs on siblings of individuals with a neurodisability. This scoping review was conducted to identify and map the characteristics of and outcomes for participants in programs designed to prepare siblings in their future roles to support their brother or sister with a neurodisability. A preliminary search on the JBI Database of Systematic Reviews and Implementation Reports, Cochrane Database of Systematic Reviews, PROSPERO, PubMed and CINAHL did not identify any reviews on this topic.

Review questions

This scoping review was conducted to answering the following two questions:

i. What are the characteristics of programs designed to support siblings of an individual with a neurodisability (e.g., purpose, description, eligibility criteria, length, activities, service provider, delivery) for TD siblings of individuals with neurodisabilities and;
ii. What are the outcomes for siblings of individuals with neurodisabilities participating in the programs?

METHODS

This scoping review was conducted according to the Joanna Briggs Institute (JBI) methodology for scoping reviews [15]. The protocol for this review has been published [16]. This report of the scoping review results was written using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist [17].

Patient and public involvement

An integrated knowledge translation approach was used in this scoping review, which is an approach to doing research with knowledge users as equal partners with researchers [18]. We partnered with the Sibling Youth Advisory Council (SibYAC) comprised of six young adults who have a sibling with a disability. The SibYAC identified the importance of the research questions addressed in this review and the program outcomes (e.g., knowledge acquisition, skill development, empowerment). The SibYAC also reviewed the preliminary findings, provided recommendations to interpret the results, and suggested knowledge translation and dissemination activities to share these results with the community.

Search strategy

An initial limited search was conducted on PsycINFO to identify relevant articles. The text words contained in the titles and abstracts of relevant articles, and the index terms to describe these relevant articles were used to develop a full search strategy for PsycINFO. The full search strategy was then adapted for each included database. The reference list of all included sources of evidence was screened for additional relevant studies. Articles published from database inception to December 20, 2020 were included.

Information sources

The databases that were searched included PsycINFO, Cumulative Index of Nursing and Allied Health Literature (CINAHL), Sociological Abstracts, Education Resources Information Center (ERIC), EMBASE, Web of Science, MEDLINE (Ovid), and Sport Discus.

Study of evidence selection

Following the search, all identified citations were collated and uploaded into Covidence, systematic review software (Veritas Health Information, Melbourne, Australia) and duplicates were removed. Following a pilot test, titles and abstracts were screened independently by two reviewers (LN, JB) against the inclusion criteria. Potentially relevant sources were retrieved in full with citation details. The full text of selected studies was assessed in detail against the inclusion criteria by two independent reviewers (LN, JB). Reasons for exclusion of full text studies were recorded. Any disagreements between the reviewers at each stage of the selection process were resolved through discussion or consultation with a third reviewer (MK).

Inclusion criteria

Participants. This review focused on identifying and describing programs with participants who are siblings of an individual with a neurodisability. For this review, neurodisability is defined as a group of congenital or acquired long-term conditions that resulted from an impairment of the brain and/or neuromuscular system and can lead to functional limitations [2]. In this review, no age limits were applied for the population of the TD siblings and siblings with a neurodisability, and may vary, including children, youth, and adults. In this review, we refer to the siblings of individuals with a neurodisability who participated in the programs as TD siblings. However, we recognize that these siblings may have had disabilities themselves that were not disclosed in the included studies.

Concept. This review included studies that described programs designed to support TD siblings in their roles. The outcomes of these programs were operationalized to include:
1) knowledge acquisition or skill development for the TD siblings themselves (e.g., knowledge about neurodisability, sharing and learning experiences about the strengths and challenges in the sibling relationship, development of coping strategies and problem-solving skills); and 2) empowerment to train TD siblings to learn skills that they can apply with their sibling with a neurodisability (e.g., how to modify certain behaviours of the sibling with a neurodisability, how to enhance social communication skills in the sibling relationship). Studies about programs that focused only on therapy or support for TD siblings without reference to support for the individual with a neurodisability have been excluded.

Context. The context of this review included all settings that deliver programs for siblings of individuals with a neurodisability, such as school, rehabilitation, healthcare, or community settings, in any country. Only studies published in English were included.

Types of sources

This review included all study designs such as experimental and quasi-experimental study designs, randomized controlled trials, non-randomized controlled trials, before and after studies and interrupted time-series studies, descriptive studies, observational studies, qualitative studies and mixed methods studies.

Data extraction

An initial pilot test of the data extraction for approximately 10% of the included studies (n=7) was performed independently by two reviewers (LN, JB) using a data extraction sheet. Based on this initial pilot test, the data extraction tool was modified to provide additional clarity of the information that would be extracted (see Supplementary File 1 for an updated data extraction sheet). For the remaining included studies, data were extracted by one reviewer (LN) and checked by a second reviewer (JB). Disagreements between the reviewers during this check of the extracted data were resolved through discussion or consultation with a third reviewer (MK). The authors of included articles were contacted to request missing or additional data based on the data extraction sheet.

Data analysis and presentation

The extracted data are presented in tabular form that provides a comprehensive overview about sibling programs based on the information outlined in the data extraction form. To address the two research questions of this review, an accompanying descriptive narrative summary is provided in this report.

RESULTS

There were 5674 non-duplicate articles retrieved through the database searches. After title and abstract screening, 5420 articles were excluded. There were 254 articles reviewed in full-text, and 196 articles did not meet the inclusion criteria. A total of 58 articles were included in this review (see Figure 1), representing 54 distinct sibling programs.

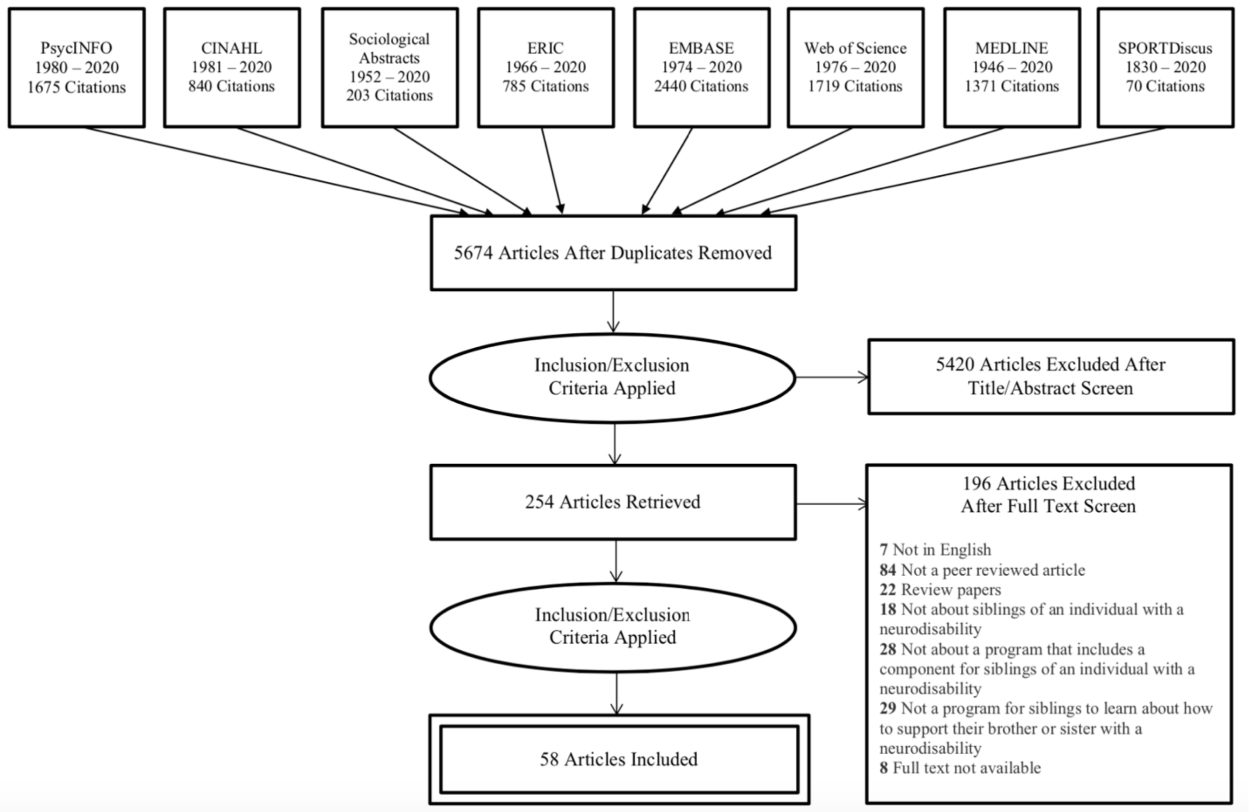

Figure 1. Preferred reporting items for systematic reviews and meta-analyses (PRISMA) diagram outlining the selection process of included studies.

Characteristics of included studies

The included articles reported studies from Canada (n=3), United States (n=42), United Kingdom (n=4), Ireland (n=1), Turkey (n=1), Norway (n=1), Sweden (n=1), Greece (n=1), Taiwan (n=1), Cambodia (n=1), and Australia (n=2). The articles reported on studies that were conducted from 1975 to 2020, with more than half of the included studies published since 2012 (n=30). Descriptions of included studies are presented in Table 1.

[insert Table 1 about here]

Participants

There was a total of 1033 TD sibling participants (n= 399 males; n=553 females). Sibling participants ranged in age from 4-67 years, with 49 studies with participants younger than 18 years old, three studies with participants 18 years or older, and two studies with a mixture of participants younger and older than 18 years of age (refer to Table 1). There were 22 studies that included participants of siblings with one neurodisability, including 18 studies that focused on autism spectrum disorder, and three studies that focused specifically on attention deficit hyperactivity order, cerebral palsy, and Down's syndrome, respectively. The remaining studies had participants of siblings with varying health conditions including intellectual and developmental disorders, or referred to disabilities as a broad term. The birth order between sibling participants and the sibling with a neurodisability was reported by 37 studies, in which 21 studies had all sibling participants who were older than the sibling with a neurodisability, two studies had sibling participants who were younger than the sibling with a neurodisability, and 12 studies that reported a combination of sibling participants who were older and younger than the sibling with a neurodisability. There were two studies that each reported two twins and one triplet. Detailed participant characteristics in each study are provided in Supplementary File 2.

There were 27 programs that included parental involvement, for example, by completing questionnaires [33,40,76,87,101,122,125,137] or they were trained to be observers of their child's performance [36]. Some parents were participants in the program [13,14,55,59,67,106,138,139,141], with parent training programs [138], information sessions [141], parent-specific sessions with some parent-sibling sessions [13,55,59,67] or as part of a family training program [14,106,129].

Concept

There were 27 programs that focused on the outcomes of knowledge acquisition or skill development by the TD siblings for themselves, and 31 programs that focused on the outcome of empowering TD siblings to be trained in specific skills that they can then teach their sibling with a neurodisability. Programs that focused on knowledge acquisition or skill development for TD siblings were first studied in the 1980s, while programs that focused on empowering TD siblings have been available since the 1970s (See Figure 2). For programs about knowledge acquisition or skill development for TD siblings, the program characteristics are presented in Table 2 and the outcomes and key findings are presented in Table 3. For programs about empowering TD siblings by training them with skills that they can teach to their sibling with a neurodisability, the program characteristics are presented in Table 4 and the outcomes and key findings are presented in Table 5.

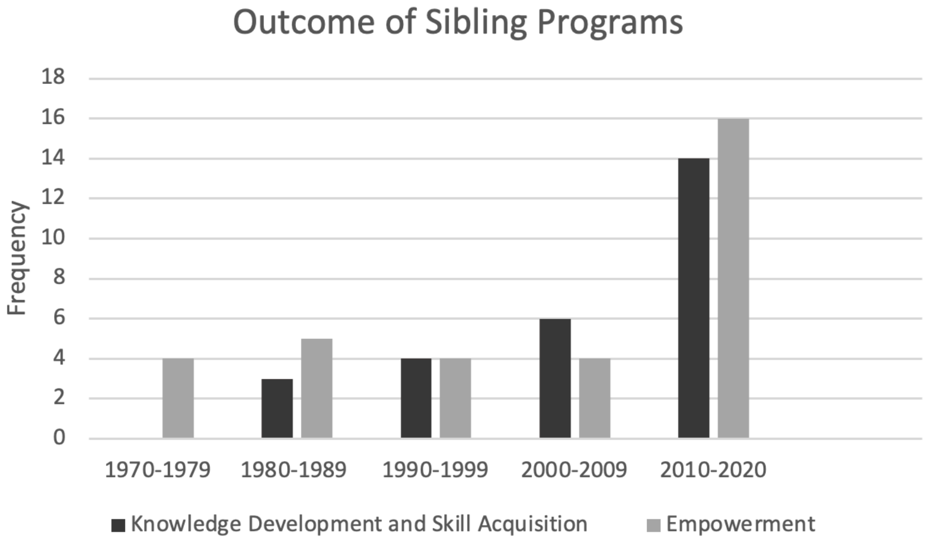

Figure 2. *The outcome of sibling programs over time.*

Across all sibling programs

Mode of delivery. Most programs were delivered in person (n=57). Only one program that offered two in-person meetings before incorporating group meetings by telephone [66].

Duration and frequency. The length of the program varied depending on the purpose of the program. For programs that were focused on providing knowledge or skills for the siblings themselves, the sessions were often offered as consecutive weekly sessions, for example, six to ten weeks for approximately half an hour to two hours [14,20,49,62,66,67,70,76,83,95,101,115,122,123,125]. There was one program that was offered for 15 weeks with 2.5-hour sessions [110]. Some programs that offered knowledge or skills acquisition for the siblings themselves in a short timeframe, for example, with all sessions in one day [55] or two days [24], or for five days as part of a summer camp [141]. The programs that trained siblings about specific skills to be applied to the relationship with the sibling with a neurodisability also varied in length. Most programs were delivered with 1-2 sessions per week, between 5 to 22 weeks with sessions ranging from 15 minutes to two hours [30,31,33,35,36,40,53,69,86,95,133]. One program described the implementation of a script-fading procedure, in which training ended when the sibling participant correctly implement each component of the procedure which was usually achieved within 30 minutes [19]. The study also described that additional booster sessions were offered if the skills were not maintained [19].

Program developers. There were 35 studies that described the program developers, with 17 programs focused on providing knowledge or skill development for siblings [11,13,14,20,24,38,59,62,66,70,76,83,101,115,122,123,139,141] and 17 studies that aimed to train siblings to apply skills with their sibling with a neurodisability [31,40,45,46,53,69,86,87,95,124,127,129,133–137]. For programs that aimed to provide knowledge or skill development for the siblings, the developers of the program were primarily from the study authors [13,20,59,83] or organizations that focused to support siblings of individuals with disabilities such as the Sibling Leadership Network in the United States [24] or Sibs in the United Kingdom [70]. There were no studies that explicitly described whether siblings of individuals with a neurodisability were part of the team of developers and the roles that they might have had. Some studies referred to elements of a program based on previous studies about sibling studies [13,76,101,139,141]. For example, a randomized controlled trial [76] that evaluated a sibling program developed the program based on a study by Kryzak and colleagues [83]. Similarly, programs that aimed to empower and train siblings to teach skills to their sibling with a neurodisability were often based on existing literature or intervention programs [31,40,46,53,69,86,87,95,127,129,133–137].

Program facilitators. There were 48 studies that described program facilitators [13,14,19,20,24,30,31,33,35,37,38,40,46,49,50,55,59,62,66,67,69,70,76,83,86,87,95,101,106–110,115,122,123,127–129,131–134,136–139,141]. The program facilitators included individuals from a variety of backgrounds including undergraduate students [31,35], graduate students [13,14,20,30,67,76,83,115,122,129,137–139], adult siblings of individuals with intellectual and developmental disabilities [24], healthcare professionals (e.g., social worker, nurse practitioners, nurses, psychologists, therapists) [33,38,50,55,62,66,86,101,106,115,122,127,139,141], community centre staff [110], special education teachers or staff members at schools [70,83,108]. For some programs, the study authors were also the program facilitators [30,37,40,46,69,87,95,132–134,137]. Some programs offered opportunities for students to be volunteers and facilitate sessions alongside licensed professionals [83,110]. While some programs required the facilitators to be licensed professionals, there were programs that also required the facilitators to receive training [55,70,83,133], such as an e-learning course for approximately 1 hour with a 2-day workshop [55] or weekly meetings with discussion, coaching, and feedback [133]. One study provided suggestions of facilitators with specific educational backgrounds that might be a good fit to run certain sessions of a program, for example, discussion sessions can be facilitated by teachers, parents, individuals from community organizations, or the siblings themselves [128].

Context

Among the programs that focused on knowledge acquisition and skill development for TD siblings, there were 16 programs that described the setting context. These programs were conducted in a variety of settings including at a community center [20,49,110], medical center [38,55,125,139] or clinic [96], school [70], or camp [141].

For programs that were focused on training the TD siblings to learn and apply skills with their siblings with a neurodisability, there were 27 programs that listed the setting context [19,30,31,33,35–37,40,45,46,53,69,75,86,87,107–109,124,127,131–138]. The majority of these programs were conducted in the participant's homes [19,30,35–37,40,46,53,69,75,86,87,107–109,124,131–137]. Some programs had sessions that were held in multiple settings. Programs were also held at a community centre [31], recreational camp [138], treatment centre [107] or behavioural management centre [127], or at a clinic [33].

Programs with outcomes of knowledge acquisition and/or skill development for the TD siblings

Purpose. The purpose of the programs was focused on the outcomes of knowledge acquisition and/or skill development for the TD siblings. To achieve these purposes, there were six programs that provided general information about the developmental or health condition [50,96,110,122,128,141]. Other programs provided information tailored to a specific condition, with four programs providing information about autism spectrum disorder [20,76,83,125]. Inherent to many programs to acquire knowledge, was a goal of creating opportunities for TD siblings to connect with peers, for example to discuss the neurodisability of their siblings with other TD siblings [55,122] or to share their lived experiences of growing up with a sibling with a neurodisability. In addition to knowledge acquisition, several programs included skill development components which included opportunities for TD siblings to develop coping skills [76,101] or problem-solving skills to enhance their relationship with their sibling with a neurodisability [20,66,76]. For example, programs offered opportunities for TD siblings to share their lived experiences and learn from each other about how to address certain situations [11,50,55,96,101].

Description of program activities. The programs included multiple sessions with a dedicated focus for each session: introductions, structured activities, and concluding session. For the introduction sessions, the content might include icebreaker activities to develop group cohesiveness and rapport [62,76,123]. After the introduction sessions, there were multiple sessions with structured activities. These activities included providing knowledge about neurodisability [49,55,59,62,66,67,83,115,122,123,139,141], learning how to problem-solve and address challenges with a sibling with a neurodisability [13,14,50,62,76,83,123]. One program included activities for adult siblings to learn about disability policy, advocacy, peer support, as well as national, state, and local resources [24]. Some of the structured activities were focused on further development of group rapport such as recreational and social activities [11,13,49,139] or arts and crafts activities [49,110,123]. For the concluding session, some programs ensured that the last session was a celebration, such as with a graduation [13,67], presentation of diplomas [66], or fun activity chosen by the TD siblings [123]. Details about the purpose and activities for each program are presented in Table 2.

[insert Table 2 about here]

Program outcomes and key findings. The programs focused on the outcomes of knowledge acquisition and skill development for the TD siblings. The following information is an overall summary about the key findings from the programs on TD siblings. The TD siblings acquired knowledge from these programs, in which they experienced an increase in understanding about disabilities after the program [11,13,20,50,62,66,70,83,96,123,125,139,141], and some TD siblings learned about new resources that they could access [24]. By participating in these programs, TD siblings identified that they found a support network because they were able to connect with other sibling participants [83,101,110,141]. The TD siblings also experienced outcomes related to the development of skills for themselves, such as development of self-esteem [50,110,141], development of coping skills, [3,49], decrease in stress [20], improvement in mood [18], and feelings of empowerment [24]. However, one study identified that the TD siblings experienced an increase in self-esteem and development of coping strategies but these outcomes were not maintained at follow-up [115]. Details about the program outcomes and key findings for each study are presented in Table 3.

[insert Table 3 about here]

Programs to empower TD siblings to teach skills with their sibling with a neurodisability

Purpose. The programs focused on empowerment by training TD siblings to learn skills that they can then teach their sibling with a neurodisability. The programs primarily trained TD siblings to learn specific skills training related to either behaviour modification skills [45,86,124,138] or development of social communication skills [19,31,33,35,46,53,69,75,127,129,131–137]. Programs that aimed to enhance social communication skills between the siblings with and without a neurodisability also had procedures, such as a behavioral skills training [129], joint attention intervention [53], milieu teaching procedures [69], natural learning paradigm [127], reciprocal training intervention [137], and script fading procedure [19]. One program aimed to train the TD sibling to be a therapist for the sibling with a neurodisability [95], while another program focused on having the TD sibling involved in behavioural treatment programs for the sibling with a neurodisability [106]. There was one program focused on addressing sibling conflict, in which both the sibling with and without a neurodisability can learn about social and emotional factors that can help to resolve conflicts [87].

Description of program activities. The programs included activities to train the TD siblings to teach their sibling with a neurodisability certain behavioural skills. The following information describes the format and content of these activities to teach these behavioural skills to the TD sibling. The TD siblings were introduced to the behavioural skill using a variety of methods including discussions with the trainer [30,31,36,53,86,87,108,109,124,129,131–134,136–138]; teaching materials such as visual text on a PowerPoint, written manual, or handouts [46,69,131,137]; completing homework sheets [33]; reading stories [134–136] or using puppets [135,136] to illustrate the skill; videos about how the sibling could prompt their sibling with a neurodisability to use the skill and how to praise their sibling for using that skill [40,46,107,109,124,127]; modeling the skill with the sibling with a neurodisability by a trainer [30,75] or parent [106] while the TD sibling observes; or having a parent explain the skill as a story to the TD sibling [134]. After the TD sibling learned the behavioural skill, some programs offered opportunities for the TD sibling to apply how they could teach the skill through verbal practice, questions, and application activities [46,138], role-play with the trainer with feedback [53,69,86,107,108,127,129,132,133,135,137], and additional prompts are provided by the trainers during the teaching sessions as needed [53,132]. Details about the purpose and activities for each study are presented in Table 4.

While all programs were focused on empowering and training the siblings, there were seven studies that reported about programs with components for other family members. Some programs offered a parent component, such as parent information sheets [115,122] or an information session [139,141]. There were three family programs, with sessions for parents and TD siblings to interact and TD siblings could share the challenges that they experienced with their parents [55,59,67]. There was a program that included both parents and TD siblings, where parents watched the videotapes that were recorded by the TD siblings, and there was a discussion about the topics discussed in the videotapes [139]. In addition to a parent component, there was also a component for the siblings with a neurodisability. Two programs that offered separate sessions that were conducted simultaneously, with a session about social communication for the siblings with autism spectrum disorder and one session for the TD sibling [76,83].

[Insert Table 4 about here]

Program outcomes and key findings. The programs focused on the outcomes of empowerment in which TD siblings could successfully carry out the skills that they were trained in [30,31,33,36,40,45,53,75,86,95,107–109,124,127,129,131,132,137]. These skills were primarily focused on the development of positive social behaviours, such as sharing, asking or giving help and compromising [40], setting and monitoring goals [86], or providing tangible reinforcement of a behaviour from the sibling with a neurodisability [53,86]. Other skills that the TD siblings learned include teaching the sibling with a neurodisability of basic self-care skills [95] or how to communicate with their sibling with a neurodisability [46]. Some TD siblings stated that the skills were easy to learn [40,127], while other siblings described that the skills were hard to learn [137]. Some studies indicated that these skills were maintained at follow-up [33,69,86,132,133]. While learning these skills, the TD siblings reported increases in self-confidence [37] or feeling enjoyment from spending time with their sibling with a neurodisability [69]. The training of TD siblings to apply certain skills were found to be associated with increased positive interactions between siblings with and without a neurodisability [106,134]. Two studies found that the skills that TD siblings learned were generalizable to other settings [86,107] or with other children with a neurodisability [109]. Details about the program outcomes and key findings are presented in Table 5.

[Insert Table 5 about here]

DISCUSSION

This review focused on programs to support TD siblings in their future roles, and these programs aimed to provide knowledge acquisition or skill development for the TD siblings themselves, or to provide training about specific skills that can be applied with the sibling with a neurodisability. In our review of online resources [147], siblings identified in blogs and interviews about the importance of first acquire knowledge about the neurodisability of their sibling before they could learn specific skills. In addition to knowledge, the TD siblings also learned about coping skills to address the challenges that they experienced in their sibling relationship. For some siblings, the programs provided a network where TD siblings could connect and share similar experiences about their relationship with their sibling with a neurodisability. A combination of supports for TD siblings that come from family, school, peers, and healthcare professionals can be helpful to enhance their ability to cope with certain situations and enhance a positive sibling relationship [148]. It is, therefore, important to consider how programs may need to foster opportunities for TD siblings to acquire knowledge about neurodisability and develop skills for themselves, such as coping skills, before providing information about how to take on future supporting roles.

In addition to knowledge and supports for TD siblings, programs also trained the TD siblings to learn specific skills to apply with their sibling with a neurodisability. Sibling training programs taught specific skills, such as how to communicate with the sibling with a neurodisability using non-verbal and verbal cues, that were similar to sibling programs described in recent systematic reviews [149,150]. These reviews focused on how siblings have been involved as a playmate, model, or instructor in interventions for children with a disability [149] or as intervention agents in programs specific to children with autism spectrum disorder [150].

This review further builds on existing literature by describing the outcome of empowerment when TD siblings learned about specific skills that they can teach to their sibling with a neurodisability. The data collected from the recent systematic reviews provided further information with evaluations of the effectiveness [149] and social validity of the programs [150].

Value of sibling programs

Meetings have been held with the SibYAC about the value of sibling programs and the relevance of the review findings to siblings and their families. The findings of this review indicated that siblings and families valued programs to support TD siblings in acquiring knowledge, developing skills for themselves (e.g., coping strategies), and being trained to apply skills with their sibling with a neurodisability (e.g., using verbal and non-verbal cues). The results from this review identified that starting in 2002, there has been an increase in the involvement of the whole family, including both parents and TD siblings, in programs. This review identified four studies, referring to two programs, that included the parents and/or caregivers as participants alongside the TD siblings [13,55,59,67]. In both programs, there were specific sessions for siblings and parents with integrated sibling-parent sessions. The activities during these sibling-parent sessions included the TD siblings creating videos that the parents viewed [13] or for the siblings to share their challenges to their parents and for parents to practice their communication skills [55,59]. When siblings are preparing for their future roles, there should be planned conversations with the whole family [151]. Despite the important roles that the TD siblings might have in the future of their sibling with a neurodisability, there are often no formalized plans [152]. Siblings have identified that there should be clear plans in place in order for them to be prepared for their future roles [152–154].

This review identified that sibling programs took place in a variety of settings depending on the purpose of the program. Most programs that were focused on empowering and training TD siblings about skills to teach their sibling with a neurodisability were conducted at home. Programs that trained the whole family was also conducted at home [14,106,129]. It is important to consider how the home environment may be a good fit to conduct certain programs that could involve supporting the TD sibling and the whole family. The person and the environment can be viewed as a bi-directional transactional process that influence each other [155]. A good fit between the person and environment can positively influence the outcomes of the programs for both the TD siblings and the siblings with a neurodisability [155]. The SibYAC shared that the environment is not only comprised of the physical home environment, but also the family context, similar to what has been identified by other researchers [156]. The TD siblings have an important role in the family and while they may not physically live in the family home at certain times [157], they should have opportunities and space to discuss roles that they would like to have.

STRENGTHS AND LIMITATIONS

A strength of this review is that there was a clear and transparent process to conduct this review, in which protocol was published prior to conducting data analysis. The published protocol and final report were written according to the JBI methodology for scoping reviews [15,158] and Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist [17]. When conducting scoping reviews, it is important that the protocol is developed a priori and that the aims are transparent and reproducible [159]. Another strength of this review is the novel contribution of mapping the availability and type of sibling programs that have been published over time. The growth in the number of available programs for siblings has significance in understanding how siblings can be involved with their sibling with a neurodisability in different ways, such as modifying certain behaviours so that both the TD sibling and sibling with a neurodisability could communicate with each other.

This review has an additional strength by providing a methodological contribution about how an integrated knowledge translation approach was used to partner with the Sibling Youth Advisory Council throughout the process of conducting this scoping review [18]. This partnership informed the relevance of the research questions, reporting of program outcomes (e.g., knowledge acquisition, skill development, and empowerment), and implications of the findings. The Involvement Matrix [160] was used as a conversation tool about roles and responsibilities, and the SibYAC members preferred to have the roles of being a listener in which they provided with information, a co-thinker in which they were asked to provide an opinion, or an advisor in which they provided (un)solicited advice [160]. Regular updates were provided to the SibYAC at each stage of the review, and the SibYAC shared their perspectives about the implications and value of sibling programs identified in this review.

One limitation of this review is that there were some articles that could not be retrieved in full text, although the corresponding authors and relevant journals were contacted up to three times. However, all non-retrievable articles were published prior to 2013 and may no longer be available. A second limitation is that some information was not reported in the articles, such as gender of participants or developers of the program. The synthesis of the findings could only be based on the information that was reported in the studies. A third limitation is that the synthesis of extracted data to answer the second question in this review about the outcomes for the siblings of individuals with a neurodisability was only provided descriptively. A fourth limitation is that only programs published in English, and there may be other existing programs for TD siblings offered in different languages.

FUTURE DIRECTIONS

There are several areas for further research that can be conducted to enhance programs for siblings of individuals with a neurodisability. Firstly, it was striking that there was limited information identified in these programs about how siblings can prepare for their future roles. While many programs in this review provided knowledge about neurodisability to TD siblings, there was only one program that described providing resources for the TD siblings [24]. The TD siblings participating in programs may wish to access additional resources for their learning. For example, a review of sibling resources was recently synthesized across children's hospitals, organizations, and treatment centres in Canada that could be shared with TD siblings [147]. Existing programs could consider how to expand the content of their programs with resources that the TD siblings may refer to. However, these programs may provide resources to siblings but did not report these resources in their publications. The reporting of these details could be included in future studies.

Secondly, this review highlighted that there were no studies that explicitly described whether siblings of an individual with a neurodisability were co-developers. However, there was one study that identified siblings of an individual with a neurodisability who were facilitators of the program [24]. Future opportunities could be provided to siblings of individuals with a neurodisability to be engaged as partners in sibling partners with different roles, such as being a co-developer or facilitator, that would be valuable and meaningful [161]. For example, siblings of an individual with a neurodisability could be co-developers and help to identify the needs and goals of the program. This review identified that there were three studies that asked about the expectations or goals of TD sibling participants for the program [20,24,115]. As co-developers, siblings of an individual with a neurodisability could inform how the expectations and goals of TD sibling participants could be included as a component of sibling programs.

Thirdly, for future sibling programs that are conducted, there could also be an exploration about the evaluations of the effectiveness and social validity of the programs. In a recent systematic review about programs for siblings of children with a disability, the effectiveness of the programs could not be determined due to the variability in the ages of participants, diagnoses of the siblings with a disability, duration of the study, content and structure of the training in the program, and reported outcomes [149]. A different systematic review of intervention programs that involved TD siblings [150] assessed the social validity, including social significance of the goals, social acceptability of the procedures, and social importance of the outcomes. Future programs could measure the social validity using similar methods.

Furthermore, future programs could be offered that involve siblings and the whole family. This review identified a recent trend to involve the whole family in programs. The SibYAC emphasized the importance of continuing to offer programs for siblings, similar to the programs that involved parents and/or caregivers identified in this review [13,55,59,67], to provide an opportunity for TD siblings and the parents and/or caregivers to have conversations about future planning. A clear and distinct focus could be identified in separate sessions for siblings and parents and/or caregivers [162], with an integrated family session that offer opportunities for the family to have clear communication about topics that are important to them.

There should also be considerations about the delivery of sibling programs using flexible approaches to program delivery. Some programs identified in this review were also conducted in the home environment, where TD siblings may feel most comfortable in. Programs can consider about how to support and/or train the TD siblings who may want to be involved but do not live in the same physical family home as the sibling with a neurodisability. Furthermore, some siblings and families may prefer alternative options to in-person approaches. There was only one program included in this review that provided a combination of in-person and telephone meetings [66], with all remaining programs that were delivered in-person. In light of the COVID-19 pandemic, some sibling programs have been adapted to be delivered online [163]. Future sibling programs could be offered in different formats, with both online and in-person approaches, in order to meet the needs of siblings of individuals with a neurodisability.

Finally, the majority of programs identified in this review were conducted in the United States. Based on a recent scoping review, there are few programs available for siblings of individuals with a neurodisability in low- and middle-income countries [164]. Although most sibling programs are available in high-income countries, a recent study identified that sibling support provider organizations in Australia, Canada, New Zealand, the United Kingdom, and the United States were operated with minimal staffing and funding [163]. While there may be programs offered in other countries besides those included in this review, these programs have not been published. Further research and allocation of funding should be considered about how to offer and publish about these programs to support siblings and the whole family of individuals with neurodisabilities across countries.

CONCLUSION

This scoping review synthesized the characteristics and outcomes of programs for siblings to support them in their future roles with their sibling with a neurodisability. This review identified that there is an increasing number of sibling programs to provide knowledge or acquisition of skills for the TD siblings themselves, as well as to train TD siblings to learn and apply specific skills with their sibling with a neurodisability. Findings from this review can inform future directions for the development and enhancement of sibling programs. Key findings from this review identified that most sibling programs are offered in-person and in the United States. There should be considerations about the delivery of these programs using flexible approaches across countries. Further research should be conducted about how siblings and the whole family of individuals with neurodisabilities can be involved, with specific consideration about tailored sessions for each family member and integrated sessions to have meaningful conversations about certain topics such as preparation for future roles that TD siblings might have.

COMPETING INTERESTS

The authors report no conflict of interest.

ACKNOWLEDGEMENTS

This review will contribute towards the doctoral degree requirements for the primary author (LN). We gratefully acknowledge our partnership with the Sibling Youth Advisory Council throughout the process of conducting this review.

AUTHORS CONTRIBUTIONS

LN, BDR, SJ, MK and JWG contributed to the conceptualisation and design of the review. LN and JB were involved as independent reviewers for title and abstract screening, as well as full text screening. MK was involved as a third reviewer for potential disagreements that arose between LN and JB. All authors were involved in the analysis, interpretation, and presentation of the key findings. LN drafted the manuscript, and all authors were involved in reviewing this manuscript and provided their final approval.

DATA AVAILABILITY STATEMENT

All data is presented as narrative text and tables to support the findings of this review.

FUNDING

This review is funded by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream – Phase 1 held by LN. The partnership with the Sibling Youth Advisory Council is supported by the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network held by LN.

Table 1. Description of included studies.

Study	**Study aim**	**Type of methodology**	**Data collection methods**	**Mean age (SD; standard deviation, range) in years**	**Diagnosis of participant's siblings**
Akers et al. 2018 [19] (United States)	To systematically replicate the results (of script fading by researchers or instructors) with siblings serving as play partners.	Adapted alternating treatment design embedded within a multiple baseline design	Parent report of statements made by the child with a neurodisability.	10 (3.27, 6-14)	Autism spectrum disorder
Brouzos et al. 2017 [20] (Greece)	To evaluate the effectiveness of a psychoeducational group program.	Randomized controlled study	Questionnaires on demographics, knowledge [21], coping and adjustment [22], and strengths and difficulties of the sibling relationship [23].	6-15	Autism spectrum disorder
Burke et al. 2020 [24] (United States)	To determine the preliminary outcomes of a leadership and support program.	Single-arm, intervention study (a study with an intervention group without a control group)	Questionnaires about participant satisfaction and changes to the program, participation in training activities [25], community or political empowerment [26], advocacy [27], motivation to impact change [28], connectedness to the disability field [29], and open-ended questions of what participants are most hoping to improve or change for individuals with disabilities and their siblings.	36 (21-67)	Intellectual and developmental disability, which includes intellectual disability, autism spectrum disorder, cerebral palsy, Down syndrome
Celiberti et al. 1993 [30] (United States)	To alter the siblings' behaviour and reinforce their interactions with their sibling with autism spectrum disorder.	Multiple baseline across skills design	Video observation recordings	8.72 (1.09, 8.16-10.25)	Autism spectrum disorder
Chu et al. 2012 [31] (Taiwan)	To examine the effects of peer- and	Non-randomized study	Observational video recordings, observations of interaction	7.33 (2.41)	Autism spectrum disorder

	sibling-assisted aquatic program on the interaction behaviors and aquatic skills in children with autism spectrum disorder.		behaviours using a checklist [32], questionnaire with open-ended and Likert questions, and anecdotal discussions.		
Clark et al. 1989 [33] (Canada)	To carry out the following objectives: a) evaluate a group sibling training program designed to enhance social interaction between autistic children and their siblings using a nondidactic problem solving approach; b) to study its effects on various types of interactive behavior; c) to monitor shifts in sibling attitudes; d) to provide social validation within the home setting; and e) to study the temporal generalization of the program.	Multiple-baseline design with follow-up assessment	Observations. Siblings completed questionnaires about home situations, children's attitudes towards children with an intellectual and/or developmental disabilities [34]. Parents completed questionnaires to assess the extent of positive interactions, conflict, and teaching between siblings at home.		Autism spectrum disorder
Coe et al. 1991 [35] (United States)	To assess and train verbal and nonverbal play responses of children with autism using siblings as primary trainers.	Multiple baseline case study.	Observational recordings written by raters about the performances of the child with a neurodisability and sibling participant	10 (1, 9-11)	Autism spectrum disorder
Colletti et al. 1977 [36] (United States)	To measure objectively the ability of siblings to effectively work with	Non-randomized study with an ABAB reversal (withdrawal) design.	Observational recordings.	11 (0.82, 10-12)	Severe neurological impairment and autism spectrum disorder

	their brothers or sisters within the home environment. To determine the effectiveness of parents as observers of their own children.				
Craft et al. 1990 [37] (United States)	To explore the effectiveness of siblings as change agents in promoting the functional status of children with cerebral palsy.	Time-series, repeated measures	Measures of change in range of motion and independence in activities of daily living.	9.58 (3.72, 4-17)	Cerebral palsy
Crouthamel 1988 [38] (United States)	To provide time-limited support groups to approximately forty siblings of developmentally delayed children; and to heighten the awareness of community professionals to the special problems and sensitivities of siblings.	Prospective narrative research design	Description of the program presented by the author.	7-13	Developmental disability
D'Arcy et al. 2005 [11] (Ireland)	To evaluate the effectiveness of a sibling program.	Pre-test/post-test design	Questionnaire to assess the sibling’s self-concept [39], as well as a pre-Sibshop interview to evaluate the sibling's knowledge and attitude with regard to their sibling's disability, and to assess their feelings towards their sibling with a disability, their contact with other siblings of children with a disability and their wish to meet them, if they discussed their	8-10	Physical or intellectual disability, or a combination of both

			sibling's disability at home, and their experience of being a sibling within the family.		
Daffner et al. 2020 [40] (United States)	To evaluate the effects of a sibling-mediated intervention on positive and negative social behaviors of young children with attention deficit hyperactivity disorder, and evaluate the implementation integrity and acceptability of this intervention among children with attention deficit hyperactivity disorder.	Non-current multiple baseline design	Video recordings and questionnaires. Parents were asked to complete questionnaires about the behaviour of the sibling with a neurodisability [41], symptoms of ADHD [42], social communications questionnaire [43], and demographic information. Parents were also asked to complete a structured diagnostic interview [44],	10.25 (1.43, 8.33-11.75)	Attention deficit hyperactivity disorder
Doleys et al. 1975 [45] (United States)	To assess the effectiveness of a response cost contingency to modify verbal behaviour.	Non-randomized pre-test post-test case study	Observational recordings in the frequency of repetitions.	19	Intellectual and developmental disorder
Douglas et al. 2018 [46] (United States)	To evaluate a sibling communication program.	Single-subject multiple probe design	Questionnaires. Parents completed questionnaires to gather information about the communication skills of children with complex communication needs [47] and how often the sibling cared for the child with complex communication needs. Questionnaires about demographics and sibling relationships were completed by both parents and siblings [48].	10.44 (3.16, 8.08-14.92)	Speech and motor delay and an emotional disability; Down syndrome; Noonan Syndrome characterized by developmental delays, hypotonia, and vision problems

Dyson 1998 [49] (Canada)	To examine a group support program for children who had a sibling with disabilities and evaluate the effects of the program on siblings.	Program evaluation	Questionnaires of open-ended questions with a forced choice format about what siblings had learned from the program, what program components they enjoyed, and what meeting times they preferred.	7.5-12	Intellectual and developmental disability, autism spectrum disorder, attention deficit disorders sensory impairment, physical disability, learning disabilities and communication disorders, developmental delay, and unspecified disability
Evans et al. 2001 [50] (United Kingdom)	To develop and evaluate sibling support groups, designed to help brothers and sisters discuss and explore their relationships with their sibling with learning disabilities.	Qualitative evaluation	Questionnaires. Siblings were asked what they felt they liked and/or disliked about the group, and to make suggestions for the future. Siblings completed tests about family relations [51], self-esteem [52], knowledge about communication and learning disabilities. Questionnaires were also sent to parents.	6-12	Learning disabilities
Ferraioli et al. 2011 [53] (United States)	To systematically replicate an established, adult-mediated intervention with typically developing sibling teachers, and evaluate the program's efficacy in teaching joint attention (JA) skills to children with autism.	Single-subject, multiple probe design	Observation video recordings with an early communication measure [54]. Interviews were conducted with siblings with questions about the quality and quantity of time spent playing with the sibling with a neurodisability.	7.33 (0.96, 6.0 – 8.33)	Autism spectrum disorder
Fjermestad et al. 2020 [55] (Cambodia)	To describe the level of mental health of sibling and the family,	Non-randomized pretest post-test design	Translated questionnaires. Both siblings and parents completed questionnaires to measure the	12.70 (2.70, 8-21)	Learning problems, intellectual disability, autism

	and family communication. To examine the change in mental health symptoms for siblings and parents, and perceived family communication. To examine the usefulness and satisfaction among siblings and parents with the intervention. To evaluate the initial feasibility of the intervention.		sibling mental health status [56]. Each parent completed a questionnaire to assess their mental health [57]. A questionnaire was completed by parents and siblings to measure family communication [58].		spectrum disorder, attention deficit hyperactivity disorder, developmental delay, or Down Syndrome
Fjermestad et al. 2020 [59] (Norway)	To promote sibling well-being.	Randomized controlled trial	Questionnaires and interview. Questionnaires about the mental health of siblings were completed by siblings and parents [56], about family communication were completed by siblings and parents [58], about quality of life by siblings and parents [60], about sibling adaptation by siblings [61], and about sibling knowledge as an interview about the phone by siblings [13].	Not applicable	Neuro-developmental disorder
Gettings et al. 2015 [62] (United Kingdom)	To explore the acceptability and feasibility of audio-conference for support groups for siblings, to discuss whether siblings can discuss their concerns through audio-conference, and to	Non-randomized pretest post-test design.	Semi-structured interview with siblings using the Siblings' Views Questionnaire. Evaluation questionnaire and follow-up interview were completed by siblings. Parents completed questionnaires about the frequency and severity	8-13	Autism spectrum disorder, ADHD, mood disorder, obsessive compulsive disorder, Down's syndrome, oppositional defiant disorder, visual impairment,

	explore the impact of support groups.		of the symptoms for the sibling with a neurodisability [63]. Siblings and parents completed questionnaires about social, emotional, and behavioural functioning of the siblings [64], and the siblings' quality of life [65].		multiple anxiety disorders or phobias
Granat et al. 2012 [66] (Sweden)	To study the effectiveness in an outpatient clinical setting of a voluntary group intervention.	Non-randomized pre-test post-test design	Questionnaires were completed by the siblings to assess knowledge about disability [13,67] and perception of sibling relationship [68].	8-12	Attention deficit hyperactivity disorder, Asperger syndome, autism spectrum disorder, physical disability or intellectual disability.
Hancock et al. 1996 [69] (United States)	To teach older siblings to use two milieu teaching procedures, modeling and mand modeling, with their younger siblings who exhibited language delays. The study examined the effects of the intervention on the older siblings' teaching behavior and on the younger siblings' language use at home during play and snack activities.	Non-randomized study	Observational data, with verbatim transcriptions.	10.67 (1.89, 8-12)	Cerebral palsy, developmental delay of unknown origin, William's syndrome
Hayden et al. 2019 [70] (United Kingdom)	To evaluate the Sibs Talk Pilot to help inform Sibs' future work with young siblings	Non-randomized study	Questionnaires. A questionnaire was completed by the teacher to measure behavioural and emotional well-being of the siblings [71,72], and the children	9.18 years (7-11)	Autism, Down syndrome, hearing impairments or chronic medical condition

			completed a questionnaire about how they feel about school [73,74].		
James et al. 1986 [75] (United States)	To evaluate a direct prompting training strategy for increasing reciprocal interactions between siblings. To evaluate whether the program would differentially affect initiations of and responses to social interactions. To assess the generalization of interaction skills from dyadic (sibling-sibling) to triadic (sibling-sibling-peer) play groups or across settings.	Multiple-baseline design.	Recordings.	6.83-8.08	Cerebral palsy and intellectual disability
Jones et al. 2020 [76] (United States)	To examine the effects of the support group for typically developing (TD) siblings of children with autism spectrum disorder.	Randomized controlled trial	Siblings completed questionnaires about mental health and adjustment [77], anxiety [78], support by responding to yes/no questions, and coping [79]. Parents completed questionnaires for each sibling about recent difficulties [80,81]. Each sibling was evaluated using a rating scale [82].	8.31 years (3.52)	Autism spectrum disorder
Kryzak et al. 2015 [83] (United States)	To evaluate the effects of the program on the siblings' knowledge about autism spectrum disorder, peer network development, and adjustment as well as	Within subject pretest-post-test design.	Questionnaires and observational video recordings. Siblings completed questionnaires about anxiety [78,84] and knowledge modified from a measure [85]. Behavioural observations of sibling interactions were conducted using video recordings.	4-14 years old	Autism spectrum disorder, which included pervasive developmental disorder not otherwise specified, autism, Asperger's,

	interactions between sibling and child with autism spectrum disorder dyads.				or autism spectrum diagnoses
Kryzak et al. 2017 [86] (United States)	To evaluate the effectiveness of combining self-management strategies with an empirically supported social skills curriculum (i.e., Stay-Play-Talk curriculum).	Multiple baseline probe design	Video recordings.	8.5 (2.60, 6-12)	Autism spectrum disorder
Lewandowski et al. 2014 [87] (United States)	To examine the effects of a modification in the procedures for administrating comic strip conversations to address sibling conflict between a child with autism spectrum disorder and his typically developing younger brother.	Case study	Questionnaires. Parents completed questionnaires, including an autism screening measure [88], assessment about social impairments that accompany autism spectrum disorder [89], an assessment of social skills [90], and a measure about the child's Theory of Mind development [91]. Both the typically developing sibling and the sibling with autism spectrum disorder completed measures about their vocabulary and word retrieval skills [92], verbal mental age [93], and grammatical contrasts such as inflections, function words, and word order [94]. Daily dairies completed by the mother to assess target behaviour. Qualitative interview completed by the parent.	6.17	Autism spectrum disorder or Asperger syndrome
Lobato et al. 1985 [95] (United States)	To assess siblings as the primary therapist	Non-randomized study	Reports from the sibling participant and agreement checker	21	Down's Syndrome

	in the acquisition of skills particularly important for improved functioning of the disabled child within the family and home (i.e., self-care and domestic skills).		(i.e., mother) about the performance of behaviour.		
Lobato 1985 [96] (United States)	To develop and evaluate a program model that could address preschool-aged siblings' needs for simple, yet factual explanations, information, and personal-emotional support.	Single-subject experimental design, a multiple-baseline-across-subject groups	Audio recordings.	5.33 (3.75-7)	Hearing loss, left hemiplegia due to a stroke, cerebral palsy, Down syndrome, and intellectual disorder
Lobato et al. 2002 [13] (United States)	To conduct a preliminary evaluation of the intervention on the primary program goals of improving sibling knowledge, sibling adjustment to chronic illness/developmental disability and siblings' sense of connectedness to other children in similar family circumstances.	Non-randomized study	Questionnaires and interview. Interview with sibling to assess knowledge of chronic illness/developmental disability. Siblings and parents completed questionnaires about the adjustment of the sibling to chronic illness/developmental disability [97], sibling connectedness, and global behavioural functioning [98].	9.8 (8-13)	Physical disabilities, autism spectrum disorders, intellectual, medical disorders or combined psychiatric and learning disorders
Lobato et al. 2005 [67] (United States)	To conduct a preliminary evaluation of a program on young siblings' knowledge of	Non-randomized study	Interviews and questionnaires. Interviews with siblings that was structured to assess knowledge of the neurodisability that the sibling had [13]. Siblings completed	5.7 (4-7)	Autism spectrum disorders including Asperger's disorder, intellectual disability, physical

	their brother or sister's condition, sense of connectedness with others in similar circumstances, and global functioning.		questionnaires about sibling connectedness, global functioning [98,99]. Parents completed questionnaires about sibling connected and program satisfaction.		disabilities such as cerebral palsy, medical disorders such as cancer, or dual psychiatric and learning disorders such as Tourette's
McCullough et al. 2011 [100] (United States)	To describe the development of a pilot program to explore the effects of a sibling support group on participants and their families.	Descriptive study	Not applicable. Reports from sibling participants and families were provided.	Not listed.	Autism spectrum disorder and intellectual disability with limited verbal abilities.
McLinden et al. 1991 [101] (United States)	To evaluate the effectiveness of a 6-week support group for siblings of individuals with an intellectual and/or developmental disability.	Randomized controlled trial	Questionnaires and interviews. The sibling completed questionnaires to describe their feelings and behaviors associated with school and home [102], evaluate the effects of works for siblings of children with disabilities [103], and assess the helpfulness of sources of social support available to siblings of children with disabilities [104]. Parents completed a questionnaire to rate the sibling’s behavior [105]. Interviews were conducted with parents.	9.17	Intellectual disability, physical disability, or multiple disabilities
Miller et al. 1976 [106] (United States)	To describe the treatment programs of two families in which siblings were involved as therapeutic agents.	Case report	Video tape recordings.	17.25 (1.92, 15-20) *Note: ages of siblings in the first case study were not provided).	Congenital facial anomaly and slow early development, autism spectrum disorder, intellectual disability
Neff et al. 2017 [107] (United States)	To evaluate the effectiveness of video	Non-randomized study	Frequency of occurrence of the target behaviours and scoring	4.67 (0.94, 4-6)	Autism spectrum disorder

	modeling as an independent teaching tool to teach typically developing siblings how to prompt and reinforce appropriate play during activities with their sibling with autism spectrum disorder. To assess whether increases in independent play occurred with the sibling with autism spectrum disorder.		whether the occurrence was appropriate or inappropriate.		
Oppenheim-Leaf et al. 2012 [108] (United States)	To examine whether the teaching interaction procedure would be effective to: teach typically developing siblings to proficiently prompt and reinforce simple social behaviors from their siblings with autism, generalize the skills that typically developing skills learned to other settings without additional prompting, lead to increase in social interactions in the sibling relationship.	Multiple-probe design	Two observers (the teacher and a reliability observer) recorded data with video recordings.	4.67 (0.47, 4-5)	Autism spectrum disorder
Özen 2015 [109] (Turkey)	To examine whether typically developing children will use the social interaction	Multiple probe design.	Video recordings.	9.67 (0.94, 9-11)	Autism spectrum disorder

	skills in the sibling training package with their siblings with autism spectrum disorder. To examine the effectiveness of sibling-delivered iPad game activities in teaching social interaction skills to siblings with autism spectrum disorder.				
Phillips 1999 [110] (United States)	To evaluate the effectiveness of a program.	Randomized controlled trial	Questionnaires. Siblings completed questionnaires about socioemotional adjustment [84], anxiety [78], self-esteem [111], perceived social support [111,112], stress [113], family functioning [114], and quality of the sibling relationship [48,68].	11.3 (9-12)	Intellectual disability
Roberts et al. 2015 [115] (Australia)	To evaluate the efficacy of SibworkS in promoting sibling wellbeing.	Randomized controlled trial	Questionnaires. Parents completed questionnaires on behalf of the typically developing siblings about their emotional and behavioural functioning [23], social support that they receive from others [116], quality of the relationship between the typically developing sibling and the sibling with a neurodisability [68], use of avoidant coping responses and approach-based responses [117], global self-esteem [118], behavioural and emotional problems [119], and whether the typically developing siblings attended any other support program. Information about family socioeconomic status was	9.3 (1.38, 7.5-12.5)	autism spectrum disorder, Angelman's syndrome, Down syndrome, Phelan-McDermid syndrome, global developmental delay, pervasive developmental disorder, intellectual disability, and optic nerve hypoplasia

			collected based on their postal code [120]. The typically developing siblings listed three things they learned in the final session of the program. Both typically developing siblings and parents completed a questionnaire about their satisfaction with the program that was adapted [121].		
Roberts et al. 2016 [122] (Australia)	To investigate factors associated with improvement in emotional and behavioural functioning following participation in the program.	Randomized controlled trial	Questionnaires. Parents provided demographic information, including age, gender, diagnosis of the sibling with a neurodisability, and relative age between the typically developing sibling and the sibling with a neurodisability. Parents completed questionnaires about emotional and behavioural functioning of the sibling with a neurodisability [119], and the emotional and behavioural functioning of the typically developing sibling [23]. The family socioeconomic status was collected based on their postal code [120]. Parents indicated whether the typically developing sibling attended any other support services.	9.14 (1.25, 7-13)	Autism spectrum disorder, Crohn's disease, epileptic encephalopathy, attention-deficit hyperactivity disorder, global developmental delay, low muscle tone, dyslexia, dyspraxia, cleft lip, anxiety, depression, oppositional defiance disorder, sensory processing disorder, dysgraphia, Angelman's syndrome, Down’s syndrome, Phelan McDermid syndrome, global developmental delay, pervasive developmental disorder, intellectual disability and optic nerve hypoplasia

Rye et al. 2018 [123] (United Kingdom)	To describe the development and qualitative evaluation of a pilot sibling group that aimed to increase siblings' understanding about disabilities, provide a space for peer support and help young people learn skills to help themselves and their siblings at difficult times. To provide a personal account of the experience of typically developing siblings who attended the group sessions.	Qualitative research	Semi-structured interviews with sibling participants.	8-13	Disabilities, including autism spectrum disorder, severe learning disability, chromosomal deletion, cerebral palsy and epilepsy
Schreibman et al. 1983 [124] (United States)	To investigate the effectiveness of a program designed to teach behavior modification procedures to normal siblings of children with autism spectrum disorder.	Multiple-baseline design	Recordings by the researchers by scoring the behavior of the sibling with autism spectrum disorder.	13, 11, 8	Autism spectrum disorder
Sheikh et al. 2019 [14] (United States)	To augment a support group for typically developing siblings of children with autism spectrum disorder with a parent intervention that focused on helping parents learn ways to	Non-randomized study	Questionnaires and observations. Parent performance of prompting and reinforcement, and TD sibling performance were observed during family play sessions. The typically developing siblings completed a questionnaire to assess the quality of their relationship with their	5 (0.82, 4-6)	Autism spectrum disorder

	support interactions between their children, one of whom has autism spectrum disorder.		sibling with a neurodisability [83] that was modified from a measure [68], and parents completed a similar measure [83].		
Smith et al. 2004 [125] (Canada)	To describe the measures and methods developed, as well results obtained, in a program evaluation of a series of sibling support groups for siblings of children with autism.	Program evaluation	Questionnaires. Parents completed a questionnaire about the internalizing (e.g., depression, anxiety) and externalizing (e.g., hyperactivity) for the typically developing sibling [98]. Typically developing siblings completed measures about how they feel about themselves [39,102], knowledge about the characteristics and causes of autism, coping and adjustment [126], and psychosocial adjustment specific to the situation of having a sibling with a developmental disorder.	10.63 (2.13, 6.58-16.25)	Autism spectrum disorder or related disorder (e.g., pervasive developmental disorder, Rett disorder, or developmental delay)
Spector et al. 2018 [127] (United States)	To determine whether siblings of children with autism spectrum disorder can learn Natural Language Paradigm and subsequently use it to occasion speech with their brothers. To assess whether children with autism spectrum disorder showed increases in language production following the	Non-concurrent multiple baseline case study design	Video recordings.	9 (1.63, 7-11)	Autism spectrum disorder and speech deficit

	introduction of sibling-mediated Natural Learning Paradigm. To assess if increases in language production generalized to untrained peers and unfamiliar settings. To measure the presence of behaviors of happiness, joint attention, and play.				
Stewart et al. 1987 [128] (United States)	Not applicable.	Descriptive paper	Not applicable.	Not applicable.	Disability.
Stewart et al. 2007 [129] (United States)	To illustrate the process by which a parent-sibling dyad was taught to implement behavioral skills training based social skill training with a sibling with autism spectrum disorder.	Case study design	Video recordings. Parent completed a questionnaire to rate the treatment and outcome [130].	10	Asperger's disorder and attention-deficit/hyperactivity disorder
Swenson-Pierce et al. 1987 [131] (United States)	To evaluate the effectiveness of a sibling training procedure designed to teach school-aged individuals with a disability to perform domestic living skills within the home environment, and to evaluate systematically the success of training procedures by	Multiple baseline design	Recordings.	12 (1.41, 10-13)	Intellectual disability, seizures, Down's syndrome, microcephaly, upper extreme spasticity

	monitoring both the use of instructional techniques by the typically developing sibling and the independent skill performance of the sibling with a disability.				
Trent et al. 2005 [132] (United States)	To evaluate procedures for teaching two responsive interaction strategies (mirroring and verbal responding) to typically developing children in the context of play sessions with their younger siblings with Down syndrome. To investigate the effects of the child-implemented responsive interaction intervention on the communicative behavior of the siblings with Down syndrome.	Multiple-baseline design across subjects and behaviors	Video recordings.	8 (1, 7-9)	Down syndrome
Trent-Stainbrook et al. 2007 [133] (United States)	To replicate a study [132] on teaching older, typically developing siblings how to use responsive interaction strategies (mirroring and verbal responding) in the context of play	Multiple baseline design.	Video recordings.	9.33 (0.47, 9-10)	Down syndrome

	sessions with their younger siblings with Down syndrome. To investigate the effects of the sibling-implemented responsive interaction intervention on the intentional communicative behavior of the children with Down syndrome. To evaluate the effects of the intervention on both the older siblings and those with Down syndrome in an untrained, generalization setting (i.e., preparing a snack).				
Tsao et al. 2006 [134] (United States)	To investigate: a) whether typically developing siblings can learn and use social skills strategies for interacting with their siblings with autism spectrum disorder and b) whether these strategies would result in increased social participation by the siblings with autism spectrum disorder.	Single-subject, multiple-baseline design.	Video recordings.	6.27 (2.83, 4.50-11.17)	Autism and Asperger syndrome
Tsao et al. 2010 [135] (United States)	Not listed. The presentation of a	Descriptive study.	Not applicable.	N/A	Disabilities

	framework for the three-step sibling-mediated social skills intervention.				
Tsao 2020 [136] (United States)	To evaluate the impact of a sibling-mediated social interaction intervention program on social behaviors of children with developmental disabilities.	Single subject, multiple probe design	Video recordings.	5.11 (1.54, 3.33-7.08)	Developmental disabilities, including autism spectrum disorder
Walton et al. 2012 [137] (United States)	To assess whether siblings correctly implemented reciprocal imitation training, whether the training had an effect on engagement with children with autism spectrum disorder, whether the skills were generalizable to other settings, whether observers were able to detect differences in pre- and post-treatment, and to measure the acceptability of the intervention.	Multiple-baseline design	Video recordings.	9.5 (1.71, 8-13)	Autism spectrum disorder
Weinrott 1974 [138] (United States)	To evaluate a training program in behavior modification for siblings of individuals with an intellectual disability. Note that	Pre-test post-test design	Questionnaires, parental reports, and recordings. Siblings completed questionnaires about behavior situations and knowledge about disability. A play session was recorded between a sibling and a camper.	between 10 and 18 years old.	Intellectual disability

	this purpose was not explicitly stated.				
Williams et al. 1997 [139] (United States)	To evaluate the outcomes of a structured, educational, and support group intervention for siblings of children with chronic illness (cancer, cystic fibrosis, diabetes, and spina bifida), including a session with parents about sibling needs; and to describe sibling and parent perceptions of sibling experiences at home.	Pretest-post-test design	Questionnaires. Siblings completed a questionnaire about the knowledge about chronic illness [140]. Parents provided an evaluation rating of the program.	8.5 years (5.57)	Cancer, cystic fibrosis, diabetes, or spina bifida
Williams et al. 2003 [141] (United States)	To assess effects of a full and partial intervention.	Randomized, three-group, repeated measures (panel) design	Questionnaires. Siblings completed measures about their knowledge about illness, social support [142], self-esteem [143], mood [140,144], attitude toward disability [140,144]. Parents completed a measure about behavior of the typically developing sibling [145,146].	11.1 years (2.2)	Cystic fibrosis, diabetes, spina bifida, cancer, and developmental disabilities (which included Down syndrome, autism, traumatic brain injury, or cerebral palsy)

Table 2. Characteristics of programs on knowledge acquisition and skill development for the typically developing (TD) siblings.

Study and Program Name	Objective(s) for siblings without a neurodisability	Duration, frequency, and context	Activities	Developers	Resources for Development	Facilitators	Conducted Evaluation? (Yes/No)
Lobato 1985 [96]	To improve the participants' understanding of developmental disabilities; to increase their recognition of the strengths of themselves, of their sibling with a disability, and of other family members; and to improve their skills in constructively expressing negative emotions associated with their unique situations.	1.5 hours once each week for 6 consecutive weeks at a children's playroom at a clinic.	A combination of training procedures typically employed in behavioral social skills training for preschool-aged children, including modeling, coaching, rehearsal, role-play, and differential feedback. Other children were encouraged to act as models and to provide specific praise and feedback to one another. Puppets, human-figure dolls, art materials, and children's literature on disabilities were used throughout the workshop to structure activities and guide discussion.	Not listed.	Not listed.	Not listed.	No

Stewart et al. 1987 [128]	The overall goal of these sessions is to provide siblings with the experiences they need to develop a perspective that portrays their handicapped brothers and sisters in a positive light.	An example of a model unit plan illustrates 45-minute sessions for 5 days.	Guest speakers, lectures, discussions, role playing, field trips, media presentations, printer materials (e.g., novels by or about individuals with disabilities, newspaper and magazine articles, songs written by exceptional individuals, commercially developed curricula, books and pamphlets), paper and pencil tasks (e.g., word puzzles, essays, research papers, poems, drawings, etc.), and picnics.	Not listed.	Not listed.	Suggestions were provided for facilitators of each activity.	No
Crouthamel 1988 [38]	To provide support to siblings.	Eight consecutive Saturdays at a regional medical center.	Introductory session for children and parents to meet. Second session to view a videotaped discussion by a group of adult siblings of individuals with disabilities. Third session to ask medical questions	Not listed.	Not listed.	A social worker and a nurse practitioner (also the sibling of a young adult with a developmental disability).	No

			to a nurse practitioner, with the use of medical books with illustrations and a model of the human brain. Remaining sessions for open conversations and planning for a newsletter that was an idea from a participant and endorsed by all participants. Final session to have a party with all family members.				
McLinden et al. 1991 [101]	To provide peer support, a forum for the expression of feelings (both positive and negative), and coping strategies for living with a sibling who has a disability	1 hour per week for 6 weeks	The group focused on developing participants' acceptance of both negative and positive feelings about their siblings. Information was provided about a wide range of disabilities. Numerous activities were utilized to provide group participants with a range of strategies to deal with peers and peer reactions to their siblings with	Not listed.	Procedures from the literature.	Two state-certified school psychologists.	Yes

			disabilities, including homework assignments to reinforce concepts taught during formal group sessions.				
Williams et al. 1997 [139] Intervention for Siblings: Experience Enhancement (ISEE)	To provide education, psychosocial and social-recreational components.	5-6 hours for the educational component, 3-4 hours for the psychosocial component, and 1.5 hours for the social and recreational component. Average number of hours spent on the sessions was nine at a medical center.	The sibling intervention consisted of structured, educational, and psychosocial, and social and recreational group sessions with siblings of children with chronic illness. A discussion session was also held with parents about issues related to sibling adjustment.	Professional expertise of each clinical nurse specialist member of the study.	Not listed.	Educational component was conducted the clinical nurse specialists of each diagnostic condition and a graduate student in pediatric nursing. Psychosocial component was conducted by experienced facilitators on the study team.	Yes
Dyson 1998 [49]	To provide support to siblings of a sibling with a neurodisability.	2-hour sessions for six Saturday afternoon workshops at a community recreation center.	Arts and crafts, learning about disabilities, group discussions and sharing of sibling experiences, and recreational and social times.	Not listed.	Not listed.	Not listed.	Yes

Phillips 1999 [110]	To alleviate the stress caused by having a sibling with a disability by providing information about, and facilitating understanding of, developmental disabilities and by creating a context that provided social support from peers and adults.	2.5 hours for 15 weeks at a community center.	Group discussions and check-ins, with a different topic relevant to siblings of an individual with a disability for each week. A variety of structured and unstructured recreational activities including computer games, videos, dance contests, theater arts, sports, gardening, and crafts. Each session included homework and tutoring.	Not listed.	Not listed.	Six team leaders who were paid community-center staff and seven volunteers who included community residents, high school students, and community center staff from other programs.	No
Evans et al. 2001 [50] Facing the Challenge	Facing the Challenge To assist siblings in developing positive strategies for living with their siblings who have learning disabilities and an associated challenging behaviour.	Three consecutive full-day sessions full days followed by six weekly evening sessions. Assessments were conducted in the participants' own home and during the group sessions.	Flashcards and video for TD siblings to learn how to explain their sibling's learning disabilities. Role-play scenarios were also used to explore the type of behaviours from their siblings with a disability and discuss how to manage these situations. Puppets and masks for how children visualized their sibling with a	Not listed.	Not listed.	Four staff who included community nurses, outreach nurses and a psychologist.	Yes

			disability. Relaxation techniques were also taught, and the TD siblings practised games they could play with their siblings. Time was set aside each week to discuss with each TD sibling about goals of the session, as well as explore their input and strengths for the group to incorporate for the following session.				
Lobato et al. 2002 [13] SibLink	To improve sibling knowledge, sibling adjustment to a chronic illness/developmental disability, and siblings' sense of connectedness to other children in similar family circumstances.	Six 90-minute group sessions conducted over a 6-8 week period	Two sessions targeted improving sibling knowledge and family information exchange. Two sessions targeted identifying and managing sibling emotions with problem-solving around challenging situations. One session focused on balancing the TD siblings' individual needs. The final session provided a review and graduation	Study authors.	Based on existing literature and original activities created by the authors.	Two doctoral level trainees in psychology or psychiatry.	Yes

			ceremony. Some activities to integrate the sibling and parent groups to enhance mutual understanding and perspective taking. For example, TD siblings created a video about their experiences that parents reviewed.				
Smith et al. 2004 [125]	To increase knowledge and understanding of autism and related developmental disorders, provide the opportunity for siblings to discuss their feelings in an accepting atmosphere, help siblings to share ways of coping with difficult situations unique to having a sibling with autism (e.g., through role playing); enhance siblings' self-concepts, and encourage siblings to have fun in a supportive environment.	Weekly sessions for eight consecutive weeks at a health center.	Information sessions on autism and related disorders and discussions relating to feelings and attitudes associated with living with a sister with a developmental disability. Exercises, games, and activities also focused on fun and promotion of group cohesion.	Not listed.	Not listed.	Not listed.	Yes

Williams et al. 2004 [141]	To provide structured teaching about the sibling's illness, psychosocial sessions, a 5-day residential summer camp, and two booster sibling sessions.	2-2.5 hour sessions at a 5-day residential camp with booster sessions at a medical center.	Educational sessions focused on medical information about the specific illness or disability. Psychosocial sessions focused on psychosocial issues, and TD siblings were encouraged to ask questions, share experiences, and talk about feelings of their sibling with a disability. Stress and ways of coping were discussed, and group games were played. Parent session focused on information exchange with nurse clinicians and other parents to enhance awareness of sibling needs. Booster sessions were provided to parents and TD siblings.	Consultation with pediatric nurse clinicians, nurse researchers, physicians, and allied health professional.	Based upon a review of the literature and consultations.	Pediatric nurse clinicians	No
D'Arcy et al. 2005 [11] Sibshops	To provide opportunities to meet other siblings, to discuss common joys and concerns,	3-hour monthly sessions on Saturdays for four	Monthly meetings, where a group of siblings come together to share information about	Not listed.	Based on a previously developed model.	Not listed.	Yes

	to learn how to handle situations commonly experienced by siblings, and to learn about the implications of the needs of siblings with a disability.	consecutive months.	their siblings with disabilities and to have fun. The morning consists of high and low energy activities, interspersed with discussion about disability and each sibling's experience.				
Lobato et al. 2005 [67] SibLink	To address sibling challenges.	Six 90-minute sessions of collateral and integrated sibling-parent groups.	The collateral sibling group activities alternated between explicitly focused on "main events" and other more social-recreational activities that implicitly addressed sibling connectedness. Two sessions targeted improving sibling knowledge and family discussions about the child's condition. Two sessions targeted identifying and managing sibling emotions with problem-solving around challenging situations typical for young siblings and one session	Not listed.	Not listed.	Two doctoral level psychology trainees.	Yes

			focused on identifying the strengths of TD siblings and balancing the TD siblings' individual needs within the family. The final session provided a review and graduation ceremony. Integrated sibling and parent groups also were conducted to enhance mutual understanding and perspective taking. Siblings created a videotape about their experiences as a brother or sister that parents reviewed. Sibling and parent groups joined for some sessions to read books about siblings and illness or disability together, to engage in interactive exercises, and a graduation.				
McCullough et al. 2011 [100]	To provide a fun and stimulating environment in which siblings could	Not listed.	Activities celebrated the special traits of participants and	Not listed.	Strengths perspective was kept in mind.	Not listed.	No

	relax and develop relationships with peers, to provide didactic information about developmental disabilities and psychosocial information about tools for coping with a special needs sibling, and to provide support for enhancing the development of a healthy self-concept and sense of efficacy.		their siblings with a neurodisability to underscore positive feelings of loyalty, self-efficacy, and self-esteem and to ensure that opportunities to express negative or ambivalent feelings were balanced with opportunities to find a positive perspective.				
Granat et al. 2012 [66]	To increase knowledge of the TD sibling about their sibling's disability and improve the ability of the TD sibling to hand their environment (e.g., answering questions from peers). To instruct TD siblings how to use problem-solving strategies in order to provide them with tools for dealing with their sibling relationship more effectively.	Two-hour sessions for six weeks.	Session focused on providing information about a particular disability, with questions, discussions, games. The sessions also included structured role-playing of problem-solving strategies. The TD siblings compiled a personal notebook comprised of lecture notes, stories, and photos of role-playing activities. Practical exercises, social interactions, and games were	Not listed.	Based on previous research about sibling programs.	Clinical staff from an outpatient habilitation centre.	Yes

			included. The final session was a festive occasion that included a presentation of diplomas and the siblings' evaluations.				
Gettings et al. 2015 [62]	To provide support to siblings, including group cohesiveness, installation of hope, psycho-education, and sharing of experiences.	One-hour sessions for eight consecutive weeks and were followed up three to six months after the last session. Sessions were in-person at a hospital school room and through audio-conferencing.	First two sessions focused on ice breaker exercises to encourage group cohesion. Session three provided TD siblings with the opportunity to gain a better understanding of their siblings' diagnoses with the opportunity to ask questions. Session four focused on matters relating to school to enable siblings to share their experiences at school including talking about friendships. The fifth session allowed siblings to share stories about recreational activities and daily family life. Session six focused on problem-solving to enhance coping	Not listed.	Adaptation from existing models.	A child psychiatrist and a clinical nurse specialist, who are both experienced clinicians.	Yes

			mechanisms for TD siblings. The last session focused on the TD siblings' thoughts about the future including their hopes for their sibling with a disability and their dreams.				
Kryzak et al. 2015 [83] Support and Skills Program	To decrease typical sibling maladjustment, increase sibling's knowledge about autism spectrum disorder, increase sibling social network, and improve sibling interactions.	2-hour sessions on seven Saturdays over a 9-week period in the fall and over a 9-week period in the spring at two university settings.	Siblings with autism spectrum disorder received individualized intervention in social, communication, and play/leisure skills, while the TD siblings participated in a support group focused on developing a network of peers who face similar family challenges, learning about autism spectrum disorder, and learning coping strategies. Then the sibling with autism spectrum disorder and their siblings attended an inclusive recreation time where they	First and last authors.	Not listed.	Licensed and credentialed professionals with experience with autism spectrum disorder treatment with student volunteers.	No

			practiced what they had learned in their separate groups. Each week covered a specific topic for sessions for the TD siblings. In the first few weeks, topics focused on learning about other group members, while the latter weeks focused on knowledge about autism spectrum disorder. Each weekly meeting began with an icebreaker game and ended with a homework assignment. This assignment helped inform parents of the activities in the support group and encouraged siblings to engage in activities with their sibling with autism spectrum disorder at home.				
Roberts et al. 2015 [115] SibworkS	To get to know each other and establish group goals, to note similarities and differences between oneself and one's sibling, as well as	2-hour sessions for 6 weeks.	Activities related to the objectives of each session.	Not listed.	Based on cognitive-behavioural therapy principles.	A paid group leader, who was a postgraduate clinical psychology student and a Provisionally	Yes

	sharing knowledge about various disabilities, to express feelings and seek social support, to use problem-solving approaches, to develop strategies for coping with stresses, including those relating to one's sibling, and to recognize one's individuality, even in terms of what one has taken away from the program.					Registered Psychologist supervised by a Clinical Psychologist. The group leader was assisted by a volunteer who had experience working with children, and, in most cases, experience in supervising outings for the siblings of children with disabilities.	
Roberts et al. 2016 [122] SibworkS	To get to know other participants and establish group goals, to recognize similarities and differences between oneself and one's sibling with special needs while sharing knowledge about different disabilities, to express feelings and seek social support, to apply problem-solving approaches, to develop strategies for coping with stresses, including those relating to	Weekly 2-hour sessions for 6 weeks.	Activities matched the intervention objectives and based on previous work that was found to be acceptable to siblings and/or families, as well as promote well-being (e.g., building the TD sibling's knowledge of disability, developing coping strategies).	Not listed.	Based on a model of family stress and adaptation.	Paid group leader, who was a postgraduate clinical psychology student and a provisionally registered psychologist. The group leader was assisted by a volunteer who had experience working with children and, in most cases, experience in the supervision of outings for	No

	one's sibling with special needs, and to recognize one's individuality, even in terms of what one has taken away from the program.					the siblings of children with special needs. Additionally, a research assistant attended all sessions in an administrative capacity	
Brouzos et al. 2017 [20]	To provide psycho-education about autism, emotional education, cognitive restructuring, training in relaxation techniques, problem-solving and social skills training and psycho-education about self-acceptance.	90-minute sessions for 8 weeks on consecutive Saturdays at an activity center for children with disabilities.	Weekly meetings, education and structured activities. Each group session consisted of four parts. In the first part, the introduction of the session's topic was followed by a discussion among the members and the leader. In the second part, the leader introduced the structured activities that the TD siblings often carried out individually or in pairs. In the third part the activity was processed. The final part included the closing of the session.	Study authors.	Not listed.	Female Master's student in a Counselling Psychology program, who had attended a postgraduate level group counseling course. She had an educational background in primary education and experience as a tutor for children with autism spectrum disorder.	No

Rye et al. 2018 [123]	To increase siblings' understanding about disabilities, provide a space for peer support and help young people learn skills to help themselves and their siblings at difficult times.	Two-hour sessions for ten weeks at the Ealing Service for Children with Additional Needs multiagency building.	Icebreakers for siblings to get to know one another. Discussion-based activities on disability to increase knowledge and understanding. Discussion-based activities on feelings to develop skills in recognising their own and others' feelings. Problem-solving activities to encourage development of coping strategies. Arts and crafts and team building games to encourage young people to work together. A final celebratory session which included a fun activity chosen by the young people.	Not listed.	Based on the structure recommended by Sibs, a national charity for people with a sibling with a disability.	The facilitators were two assistant psychologists who also received sibling group leader training from the organization.	No
Hayden et al. 2019 [70] Sibs Talk	To improve siblings' well-being and their engagement with learning	25-35 minute sessions for ten sessions during a school term at school.	TD siblings were guided through an activity page in a booklet. The first session started with sharing basic information about each sibling's family and circumstances to	Sibs, a charity in the United Kingdom	Not listed.	Staff members who attend a two-hour training session at their own school or at a host school nearby. The training focused on the micro	Yes

			help the sibling develop trust and rapport with the staff member leading the intervention. As the booklet progressed, the sessions focus in more depth on their brother or sisters' disability or condition, the TD siblings' feelings and experiences, and the issues that were challenging for them at home and school. The sessions also looked at the skills, knowledge and attributes that TD siblings have acquired and how their school can support them.			skills required for listening to and acknowledging siblings' feelings. Staff could contact Sibs staff for further advice if required.	
Sheikh et al. 2019 [14]	The sibling support program focused on learning about autism spectrum disorder, sharing feelings, learning coping skills, problem solving, advocacy, and creating a peer support network. The parent-sibling training focused on	1.5 hour support group session for ten weeks with 30-45 minute parent-sibling training for three to six weeks at a university.	For the support group, games and activities related to a specific topic and goal. Each session began with some warm-up games while the group waited for all the siblings to arrive. Then the leader introduced the main activity for that	Not listed.	Based on the curriculum of other sibling support groups.	First year graduate students pursuing a Master’s degree in Applied Behavior Analysis, each with over 2 years of experience working with children with	No

	increasing prosocial behaviour directed toward the sibling with autism spectrum disorder.		week, spending the majority of the time on that activity, followed by a short snack. During the first few weeks, activities and discussions focused on learning about other group members to develop group rapport and a peer network and learn about group members' similarities and differences and begin to share their feelings about their siblings with autism spectrum disorder. TD siblings took home weekly challenges to practice something at home or talk to their parents .This helped inform parents of the activities in the support group and encouraged siblings to engage in activities with their sibling with autism spectrum disorder at home.			autism spectrum disorder, and one student completing a . Bachelor's degree in Psychology. developmental disabilities and behavior therapists working with children with autism spectrum disorder and families.	

			Immediately after a parent-intervention session, the parent, TD sibling, and sibling with autism spectrum disorder participated in family play sessions.				
Burke et al. 2020 [24] Sibling Ambassadors Program	To increase awareness of the sibling experience and sibling needs among siblings of individuals with intellectual and developmental disabilities and the public more generally.	15 hours for two days.	Education, in which TD learned about disability policy, advocacy, and peer support. Training included: didactic instruction, small group activities, skill-building sessions, individual reflections, and time for siblings to socialize with one another. At the end of the training, participants created an action plan with targeted activities to increase awareness of the sibling experience in their communities.	The Sibling Leadership Network.	Based on literature.	Adult siblings of individuals with intellectual and developmental disabilities; these siblings were affiliated with the Sibling Leadership Network.	No
Fjermestad et al. 2020 [55] SIBS intervention	To provide siblings with an opportunity to discuss the disorder of their brother/sister, and associated emotions	Five sessions in one day at a mental health clinic for children and adolescents.	Sessions 1, 2, and 4 are parallel (separate) group sessions for siblings and parents. Sessions 3	Not listed.	Not listed.	Group leaders were clinical staff (i.e., psychologists, social workers, psychiatrists,	Yes

	and family challenges.		and 5 are integrated sibling-parent dialogues in which each sibling and parent talk together in pairs. Session 1 was an introduction. Sessions 2–3 focused on siblings' disorder knowledge based on a semi-structured group interview. Sessions 4–5 focused on siblings' emotional experiences based on cognitive-behavioral principles about thoughts and behavior influence emotions. In Sessions 2 and 4, siblings prepared lists of questions about the disorder and experienced family challenges, respectively, which a group leader brings to the parent group for discussion. In Sessions 3 and 5, each sibling discussed questions			and art therapist) with training to present the intervention.	

			and challenges with their parent.				
Fjermestad et al. 2020 [59] SIBS intervention	To improve their mental health and well-being.	5 sessions delivered over 2 days, 1 week apart. Day 1 comprised of sessions 1-3 (3.0 h including breaks). Day 2 comprised of sessions 4-5 (2.5 h including breaks) at municipal and specialist health service sites.	Sessions 1, 2, and 4 are parallel (separate) group sessions for siblings and parents. Sessions 3 and 5 are integrated sibling-parent dialogs in which each sibling and parent talk together, separate from other participants. Sessions primarily focused on disorder knowledge and emotional experiences, in relation to the overarching component, family communication.	Study authors developed the manual.	Not listed.	Group leaders with employment in municipal or specialist health services, health professional training, completion of a training package. All group leaders will be invited to supervision webinars 2-3 times per year.	No
Jones et al. 2020 [76]	To provide support to siblings, including a discussion of feelings related to the sibling with autism spectrum disorder and family, problem-solving and coping skills, peer network, or information about autism spectrum disorder.	2-hour sessions for 10 Saturday or Sunday mornings.	During the first hour, siblings with autism spectrum disorder received individualized instruction in social, communication, and skills while TD siblings participated in either the support group or attention-only control group.	Not listed.	Based on sibling programs previously described in the literature.	A graduate student with the assistance of another graduate or advanced undergraduate student (trained and supervised by the first and last authors), with a psychology background and	No

			During the second hour, all siblings had inclusive recreation activities (e.g., stretches, relay races). Lessons and activities focused on characteristics of autism spectrum disorder, different ways to cope with frustrations with siblings with autism spectrum disorder (e.g., take a deep breath, tell a parent) and who to go to for help. TD siblings had weekly challenges to practice something at home or talk to their parents. This helped inform parents of the activities in the support group and encouraged siblings to engage in activities with their sibling with autism spectrum disorder at home.			experience implementing the program.	

Table 3. Outcomes of programs focused on knowledge acquisition and skill development, and key findings for the typically developing (TD) siblings.

Study	Program Outcomes	Key findings for the TD siblings
Lobato 1985 [96]	Knowledge acquisition and skill development	Overall, participating in the workshop program led to positive changes in statements of self-descriptions and increased accuracy in describing disabilities. No negative side effects in behaviour. Parents reported satisfaction to have conversations with their child.
Stewart et al. 1987 [128]	Knowledge acquisition	Not applicable.
Crouthamel 1988 [38]	The author shared about the siblings' perspective from participating in the program	Enjoyment in meeting other siblings of individuals with disabilities and development of awareness of disabilities.
McLinden et al. 1991 [101]	Knowledge acquisition, skill development, and satisfaction with the program	Limited evidence of program effectiveness on knowledge, attitudes, self-concept, or problem behaviour. Reported enjoyment and higher levels of social support. Parents reported improvements in behaviour towards the sibling with disability.
Williams et al. 1997 [139]	Knowledge acquisition	A common theme found was that of siblings feeling physically and emotionally isolated from their parents, Thus, the results provide evidence that the family is a system of interrelated individuals, and that each member affects (and is affected by) other members of the system. The study intervention showed positive effects on siblings of a sibling with a chronic health condition, based on the results of the knowledge test. In addition, the parents validated the usefulness of the program for the siblings in both their numerical rating of it and their verbatim descriptions of its specific benefits to siblings.
Dyson 1998 [49]	Knowledge acquisition	Significant increase in learning, such as ways to improve relationships with the sibling with a neurodisability, and understanding about and empathy for people with disabilities.
Phillips 1999 [110]	Knowledge acquisition and social support.	Decrease in sibling-related stress, increased social support from peers and staff, less depression and anxiety, and higher self-esteem.
Evans et al. 2001 [50]	Satisfaction with the program, knowledge acquisition and skill development.	Development of self-esteem, knowledge about the needs of the sibling with a neurodisability, improvement in relationships among family members, receptivity to supporting therapeutic intervention s at home.
Lobato et al. 2002 [13]	Knowledge acquisition and skill development.	Increased knowledge and sibling connectedness. Decreased behavioural problems from sibling reports, and decreased behavioural problems from parent reports. No negative emotional or behavioural outcomes reported.
Smith et al. 2004 [125]	Knowledge acquisition and skill development	Greater knowledge of autism spectrum disorder and better self-concepts (i.e., how they feel about themselves). Enjoyment of experiments reported from participating in the program.

Williams et al. 2004 [141]	Knowledge acquisition and skill development	Improvements in outcomes of sibling mood, attitudes toward illness and toward the sibling with a neurodisability, social support, and self-esteem, as well as decreases in reported sibling behaviour problems.
D'Arcy et al. 2005 [11]	Knowledge acquisition and sharing of experiences.	Achievement of program goals. 81% of siblings wish to meet again after the program. Reported enjoyment, consistent attendance rates, able to articulate difficulties and challenging situations. Opportunity to learn something new. Ability to describe disability in terms of a social model. Program benefits were observed by TD siblings and parents.
Lobato et al. 2005 [67]	Knowledge acquisition and skill development	Increased knowledge of disability, sense of connectedness, and perceptions of self-competence. No negative emotional or behavioural outcomes observed.
McCullough et al. 2011 [100]	Although not measured, the goals of the program were on knowledge acquisition and skill development.	The group modality provided a rich resource for siblings of children with developmental disabilities.
Granat et al. 2012 [66]	Knowledge acquisition and skill development (i.e., problem-solving skills).	Ability to explain the neurodisability of their sibling increased. Less time spend with their sibling with a neurodisability, but had more fun when together. For siblings of a sibling with autism spectrum disorder, there were no significant changes in problem-solving strategies but there was increased admiration for their sibling. For siblings of a sibling with intellectual disabilities, there was an increase in the subscale Dominance, but also greater affection for their sibling.
Gettings et al. 2015 [62]	Knowledge acquisition	Provided descriptions of behavioural difficulties of their sibling with a neurodisability that was consistent with parent reports. Increased number and range of people siblings spoke to about their concerns, for example, another sibling support group member. Increased feelings of preparation due to a better understanding of the neurodisability.
Kryzak et al. 2015 [83]	Knowledge acquisition and skill developing (i.e., coping strategies)	Significant improvements in the siblings' adjustment and peer network as well as some changes in knowledge about autism spectrum disorder and interactions with the sibling with autism spectrum disorder. Decreases in depression, anxiety, negative self-esteem, and interpersonal problems.
Roberts et al. 2015 [115]	Knowledge acquisition, skill development, satisfaction with the program	Improved emotional and behavioral functioning immediately after the program and maintained after 3 months. Enhanced self-esteem and less avoidant coping, but was not maintained at follow-up.
Roberts et al. 2016 [122]	Knowledge acquisition and skill development	Greater reduction in emotional and behavioural problems if the TD siblings had greater emotional and behavioural problems at baseline and were from families where the sibling with a neurodisability had more severe symptoms, which were maintained at 3 months post-intervention.
Brouzos et al. 2017 [20]	Knowledge acquisition, skill development	Significant increase in understanding about autism spectrum disorder. Reduction of adjustment difficulties and emotional/behavioural problems. Younger participants were more likely to

		acquire valid information about autism spectrum disorder, whereas older participants were more likely to experience a decrease in their coping/adjustment difficulties.
Rye et al. 2018 [123]	Knowledge acquisition and skill development	Five themes identified: meeting similar people, a break from home, enjoyment of the activities-led group, building confidence and self-esteem, and learning and applying knowledge about disabilities. The sibling group offered young people a chance to meet with others in a similar position and for some this was the first time they had knowingly done so. The program also served to increase knowledge about disabilities and teach coping strategies the young people could use at difficult times, and some felt it did equip them with more knowledge about their sibling's disability.
Hayden et al. 2019 [70]	Knowledge acquisition and skill development	Improvements on the Strengths and Difficulties Questionnaire that measures behavioural and emotional well-being, specifically there were statistically significant changes on hyperactivity scores, prosocial behaviours, and emotional problems. Some changes in how TD siblings feel about school, conduct problems, and peer problems but were not statistically significant. TD siblings indicated that they learned new things about disability more generally. They could name individuals who they could talk to about sibling issues. Mostly positive comments about sibling relationships, although there were a few negative comments such as experiences of physical aggression from their sibling with a disability.
Sheikh et al. 2019 [14]	Knowledge acquisition and skill development	Increase in prosocial behaviours, and one TD sibling reported improvements in the sibling relationship.
Burke et al. 2020 [24]	Participation in training activities, community or political empowerment, advocacy, motivation to impact change, connectedness to the disability field	The program positively correlated with the intended outcomes of connectedness and empowerment, although these outcomes were not maintained at follow-up. TD siblings developed solidarity with other siblings, felt empowered, and learned about new resources.
Fjermestad et al. 2020 [55]	Knowledge acquisition and satisfaction with the program.	Self-reported mental health remained unchanged. High satisfaction from TD siblings and parents.
Fjermestad et al. 2020 [59]	Knowledge acquisition	Not applicable.
Jones et al. 2020 [76]	Knowledge acquisition and skill development (i.e., problem-solving and coping skills)	Significant improvements in externalizing behaviour and coping skills when compared to the control group. When the siblings with autism spectrum disorder had more severe symptoms, the program appeared to provide a buffer again symptoms of anxiety and depression for the TD siblings. No changes in support network. Older siblings were found to be helpful during discussions with the younger siblings.

Table 4. Characteristics of programs focused on empowerment by training typically developing (TD) siblings to teach skills to their sibling with a neurodisability.

Study and Program Name	**Objective(s) for siblings with a neurodisability**	**Objective(s) for siblings without a neurodisability**	**Duration, frequency, and context**	**Activities**	**Developers**	**Resources for Development**	**Facilitators**	**Conducted Evaluation? (Yes/No)**
Weinrott et al. 1974 [138] Sibling Training Program	Not listed.	To providing behavioral modification training strategies to siblings of a sibling with an intellectual disability.	7 weeks at Camp Freedom.	On the first four days at camp, each TD sibling created their own schedule based on the guidelines from the program directors: each sibling would observe three activities (classes) of their choosing and attend two group meetings. On the third day, the emphasis shifted from observation to guided instruction and interaction with campers. TD siblings were no longer watching, but were teaching. Methods of dealing with resistance and aggression were also demonstrated. Siblings were given practice working with one or two campers under direct guidance consisting of specific instructions and prompts.	Not listed.	Not listed.	Two graduate students.	Yes

				Classes during the third day usually ended with the sibling working on a one-to-one basis with their camper sibling. Sessions included information about speech (e.g., developing expressive language), reading and readiness (e.g., how to conduct a teaching session at home), creative dramatics (e.g., play skills), and intellectual disabilities including medical considerations. There were daily swimming periods and recreational activity for which they would leave camp. These events included horseback riding, bowling, seeing a movie, or going out for a pizza.				
Doleys et al. 1975 [45]	To reduce the number of verbal repetitions.	To implement a behavior modification program using a	2 weeks for intervention.	Not listed.	19 year old TD sibling, a student in an undergraduate course in	Not listed.	Not listed.	No

		response cost contingency.			the experimental analysis of behaviour.			
Miller et al. 1976 [106]	Case 1. To facilitate development of the sibling with a neurodisability by shaping her speech and encouraging her to take risks and to play alone for as long as 45 minutes. Case 2. One of the behaviour selected for intervention was to reduce the taking of food from the refrigerator for the sibling with a neurodisability.	To train siblings to be involved in behavioural treatments for their sibling with a neurodisability, and to increase positive interactions between siblings.	Not listed.	Case 1. The siblings were brought into the program; they observed the mother playing with Sally and she explained social learning principles to them in the therapist's presence. Case 2. During the first two sessions, social learning principles were taught to the family through didactic materials and role playing. A schedule was made so that each sibling had 2 regular days "on call" during which he or she might have to help with the sibling with a neurodisability. On other days they were free to plan their own activities. In addition, each sibling was asked to spend 15 minutes a day in a one-to-one activated related to	Not listed.	Not listed.	Therapist	No

				the interests of the sibling with a neurodisability, such as playing with cars or going for a walk. During these sessions they were to encourage eye contact, coherent speech, and appropriate social behaviors.				
Colletti et al. 1977 [36]	Not listed.	To train siblings as behaviour modification aides.	20-minute sessions for six baseline sessions followed by six intervention sessions at home.	Experiment 1: The TD sibling was asked to carry out a cue from the experimenter, food reinforcement, and praise. The TD sibling was told to refrain from physical or further verbal interaction with her sister. For example, the TD sibling asked her sister with a neurodisability to string the bead, receiving a cue to do so from the experimenter every 60 seconds. Experiment 2: The TD siblings were asked to carry out a task with their sibling with a	Not listed.	Not listed.	Not listed.	No

				neurodisability. When each sibling reached his first intervention phase, he was told that he would now be allowed to give the target child candy for doing the task correctly. The sibling with a neurodisability was asked by the sibling to print letters on paper with crayon.				
Schreibman et al. 1983 [124]	To learn a task or behaviour, and respond appropriately to prompts.	To provide siblings with a set of generalizable behavior modification skills, which they could use in a variety of settings with a variety of target behaviors.	Approximately 30 minutes for each session, a total of 8 sessions at home.	During the first training session the sibling and trainer viewed a videotape which presented examples of behavior therapy with autistic children. Reinforcement, shaping, chaining, and discrete trial techniques were discussed. Next, the trainer and sibling discussed how behavior modification procedures could be applied to everyday situations involving problem behaviors, using examples from a training	Not listed.	Based on literature.	Trainer.	No

				manual. The next training step involved instruction to the siblings while they worked with their sibling with autism spectrum disorder. A specific target behavior was chosen and worked on for approximately 30 min. The trainer would periodically interrupt briefly with corrective or positive feedback on the sibling's progress. If the sibling had trouble applying a particular procedure, the trainer modeled the procedure and asked the sibling to try again until he or she could perform it correctly.				
Lobato et al. 1985 [95]	To acquire skills for improved functioning (i.e., self-care and domestic skills), specifically the goals selected were	To train the sibling as a primary therapist for the sibling with a neurodisability.	Weekly meetings for five consecutive weeks. Meetings every three weeks throughout the	During the training sessions, the sibling participant was required to demonstrate mastery of the principles, application, and evaluation of behavior analysis	Not listed.	Based on literature.	First author.	No

	independent toothbrushing and bedmaking.		remainder of the project.	using quizzes and workshops. As the final step in her training, the TD sibling developed a proposal of the instructional and evaluation methods she would use for the project.				
James et al. 1986 [75]	To increase reciprocal interactions between siblings.	To train siblings to increase interactions with their sibling with a disability.	Session ranged from 12-15 minutes. Average of 5 days per week, no more than two training sessions on a single day, with 4-6 hours separating any two sessions. Total number of sessions ranged from 4-8 at home.	Modeling session to demonstrate how to initiate interactions, prompt responses, and reinforce both initiations and responses. The TD siblings practiced these skills with feedback and prompts.	Not listed.	Not listed.	Experimenters	No

Swenson-Pierce et al. 1987 [131]	To improve independent performance of domestic tasks.	To learn and effectively use instructional techniques to increase prompting and social praise in teaching a domestic task to their sibling with a neurodisability.	Sessions for 2-3 times per week, for approximately 3 hours. The initial training session was completed within an hour. All sessions were at home.	The training procedure for preparing the TD siblings to act as instructors for their brother or sister with a disability consisted of the following components: (a) an explanation of their role as an instructional agent, (b) an overview and discussion of the system of increased prompting and social praise, (c) an overview and discussion of the task analysis, (d) a period of role playing where the instructor portrayed as the sibling with a neurodisability, and (e) a period of in vivo instruction accompanied by instructor feedback.	Not listed.	Not listed.	Instructor.	Yes
Clark et al. 1989 [33]	To enhance social interaction between siblings with and without autism spectrum disorder.	To train siblings to enhance their social interaction with their sibling with autism spectrum disorder.	Nineteen 2-hour sessions at clinic.	During baseline, siblings were taught a general problem-solving strategy to introduce the children to role playing and problem-solving discussion format.	Not listed.	Not listed.	Therapist.	No

				The children were asked to generate alternative solutions and to evaluate their potential effectiveness. The therapists then role played these ideas. Siblings were then prompted to evaluate their suggestions and to practice these strategies in role playing with the therapist.				
Craft et al. 1990 [37]	Not listed.	To promote spontaneous activity among siblings, which could help children with cerebral palsy.	Twice a month, for four months at home.	Two phases: educational and reinforcement of learning. Educational content was divided into discussions of cerebral palsy (e.g., “What is cerebral palsy?”) and promoting motivational skills (“What can I do to help my brother or sister be more independent?”). Learning reinforcement activities focused on ways to increase the functional skills of the siblings with cerebral palsy.	Not listed.	Not listed.	Study investigators	No

				Activities for recreational and for the siblings to share feelings and problems were included.				
Coe et al. 1991 [35]	To target behaviours functional manipulation of play materials and activity related verbalizations (i.e., verbal requests for play materials, compliments to siblings, comments or descriptions of play activity).	To target behaviours, including appropriate use of verbal prompts, nonverbal prompts, verbal reinforcement and tangible reinforcement to shape behaviour.	Treatment sessions began with a five-minute training period. Sessions were conducted for 2 to 3 afternoons a week for approximately 15-20 weeks. All sessions were conducted at home.	Training of: 1) nonverbal play behaviour (assembling Tinker Toy pieces or rolling a truck, depending on the child-sibling pair) and 2) verbal behaviour (requesting the Tinker toy piece or the truck). Nonverbal prompts taught to the sibling include 1) use of manual guidance of the head to establish attention; 2) guidance of hands in executing nonverbal play responses; 3) response interruption of stereotypes and inappropriate responses such as throwing toys or stealing reinforcers. Verbal prompts taught included: 10	Not listed.	Not listed.	Trainer and undergraduate students.	No

				activity orientation ("We're going to play with the tinker toys (truck) today and earn stickers and chips"); 2) direction and behaviour ("Say 'Stick please!'"; "Say 'I want the truck, please' for a chip;". Reinforcement training encompassed delivery of both verbal praise and tangible reinforcers (e.g., sticker or edible).				
Celiberti et al.1993 [30]	Not applicable.	To provide training of a generalized set of behavioural skills to siblings.	15-minute training session for 13-17 sessions at home.	The training curriculum included: how to deliver play-related commands effectively, how to use social praise for appropriate responses, and how to respond when the child with autism did not comply with a request. The trainer demonstrated the behavioral skills for a maximum of 5 minutes with the sibling with autism	Not listed.	Not listed.	The first author, a graduate student in clinical psychology with over two years of intensive experience with children with autism spectrum disorder.	Yes

				spectrum disorder while the TD sibling observed. The trainer discussed the various skills and procedures to be acquired. For the next 10 minutes the sibling practiced the behavior with the child with autism, while the trainer offered feedback. If necessary, the sibling role-played with the trainer behaviors that were causing difficulty. The sibling was trained successively in each of the three sets of skills until she reached the criterion of 80% correct responding over two consecutive sessions for each set.				
Hancock et al. 1996 [69]	To improve the siblings' language use at home during play and snack activities. Child language behaviors consisted of (a)	To train siblings to teach two milieu teaching procedures, modeling and mand modeling.	45-minute training sessions, for a total of four sessions at home.	Training consisted of: presenting the sibling with a written manual that had been designed specifically to fit each sibling's interests and reading	Not listed.	Modification of training package previously used with parents.	First author, a female with masters-level preparation in child development and 3 years experience implementing	No

	frequency of total child utterances, (b) frequency of targets used spontaneously, and (c) frequency of total use of targets (prompted plus spontaneous). Table 2 provides definitions.			comprehension level (assessed informally based on grade level), discussing the material presented in the manual with the sibling, watching videotapes modeling the milieu teaching techniques with children who exhibited language delays, watching videotapes of the TD sibling and sibling with a neurodisability recorded during baseline and having the sibling identify appropriate instances for applying the milieu technique, role playing use of the milieu techniques with the adult trainer, and engaging in practice sessions with the TD sibling and sibling with a neurodisability with feedback.			milieu teaching procedures with young children.	

Trent et al. 2005 [132]	To improve the communication behaviour.	To teach two responsive interaction strategies (mirroring and verbal responding).	30–60-minute sessions, two sessions per week, with each, approximately 35 sessions in total at home.	The investigator taught the TD sibling one of the responsive interaction strategies or reviewed the previously taught strategies. The teaching and reviewing portion of the intervention consisted of five components: presentation of information by the investigator with a manual, opportunity for the TD sibling to discuss the procedures and ask questions, use of modeling and role play to practice using the strategies, a second opportunity to discuss and ask questions, and setting up the activity for the play session.	Not listed.	Not listed.	Investigator and trainer	No
Tsao et al. 2006 [134]	To increase social participation.	To learn and use social skills strategies for interacting with their siblings with autism	About 10 sessions, for 10 minutes each at home.	Lessons taught common behavioral strategies for facilitating social interactions with the siblings with autism spectrum	Not listed.	Based on literature.	Researcher (the first author)	Yes

		spectrum disorder.		disorder, such as establishing eye contact, suggesting play activities, initiating conversations, or offering or asking for help. The training involved the researcher reviewing the previous day's lesson, introducing the new skill, and reading a story to illustrate the use of the skill. The researcher modeled examples of new behaviours and parents could help to explain the story and behaviour.				
Stewart et al. 2007 [129] Behavioural skills training	To improve conversation skills, with special emphasis on proper eye contact, soliciting input from the conversational partner regarding his or her interest in the topic, and avoiding topics on which the	To learn how to implement behaviour skills training to teach social skills.	60 sessions.	Behaviour skills training was provided, comprised of instructions, modeling, rehearsal, and feedback. Instructions included a rationale for treatment, a description of the treatment process, and a clear presentation of the target behaviour. The target	Not listed.	Literature about behavioral skills training and principles of behaviour modification.	Graduate student therapist (first author)	No

	sibling with autism spectrum disorder typically perseverated.			behaviour was modeled several times during various practice scenarios, followed by opportunities to perform the target behaviour during various rehearsal scenarios with feedback on the performance.				
Trent-Stainbrook et al. 2007 [133]	To improve the intentional communicative behavior.	To teach two responsive interaction strategies (mirroring and verbal responding).	30–60-minute sessions, two sessions per week, 19 sessions in total at home.	Intervention sessions were divided into three segments during both conditions of intervention. The first segment included either the trainer teaching the older TD sibling one of the responsive interaction strategies or reviewing the previously taught strategies. There were four subcomponents to the teaching and reviewing portion of the intervention: (a) presentation of information by the trainer with the use of a Responsive Interaction Pictorial	Not listed.	Adapted from previous studies.	First author and research assistant.	No

				Manual developed for this project and available from the first author, (b) opportunity for the TD sibling to discuss the procedures and ask questions, (c) use of modeling and role play to practice using the strategies, and (d) a second opportunity to discuss and ask questions.				
Tsao et al. 2010 [135] Three-Step Sibling-Mediated Social Skills Intervention	To increase interactions between typically developing siblings and their sibling with a disability.	To learn strategies to elicit social interactions from their sibling with a disability, and learn ways to help their sibling with a disability become involved in play or social interactions.	Frequency and duration of intervention was not listed. The intervention was conducted at home and in the community.	The first one or two basic lessons help siblings learn how to use strategies consistently to elicit social interactions from their sibling with a disability. The subsequent lessons provide TD siblings some specific ways to help their brother or sister with a disability become involved in play or social interactions.	Not listed.	Based on a model.	Not listed.	No

Ferraioli et al. 2011 [53] Joint Attention Intervention	To learn joint attention skills, which is defined as the ability to use "gestures and eye contact to coordinate attention with another person in order to share the experience of an interesting object or event".	To train siblings to implement a joint attention intervention with their sibling with autism spectrum disorder.	15-minute training sessions two or three times per day, 1-2 times per week. Sessions ranged from 7-9 weeks at home.	Procedures of the intervention were reviewed with the TD sibling. The TD sibling then participated in a brief interactive instruction with the experimenter, including modeling and role-plays with feedback provided. Prompts were provided to the TD sibling when needed.	Not listed.	Based on previously described procedures from the literature.	Not listed.	Yes
Chu et al. 2012 [31]	To increase aquatic and social skills of both children with autism spectrum disorder and TD children.	To provide training to typically developing peers and siblings of individuals with autism spectrum disorder.	60-minute sessions for 32 sessions in total over 16 weeks at a local indoor hydro-therapy and swimming pool.	A workshop was held to introduce the aquatic program, and TD siblings were completed a training course. Rules and roles were discussed. Assisting steps were introduced: (a) physical interactions and (b) social interactions. The program consisted of warm-up activities, teacher instructions, group games, and cool down activities in each group, including the	Not listed.	Based on literature.	Instructors had under-graduate degrees in physical education, 1-2 years of experience working with individuals with autism spectrum disorder, and an additional training course by the primary researcher.	No

				sibling-assisted group.				
Oppenheim-Leaf et al. 2012 [108]	To increase play interactions between siblings.	To teach how to get their sibling with a neurodisability to play with them, how to get their sibling with a neurodisability to share toys with them, how to provide play-related instructions, and how to find out what the sibling with a neurodisability wanted to play.	20-30 minutes of teaching periods for 40 sessions at home.	Sessions during the teaching phase always began with role-play and generalization probes for the current skill. Once all probes of the day were completed, the TD sibling was taught the current target skill through the use of the teaching interaction procedure. The teaching interaction procedure involved didactic teaching, modeling, and role-plays.	Not listed.	Not listed.	Teacher.	No
Walton et al. 2012 [137] Reciprocal Intervention Training	To increase reciprocal imitation skills in a naturalistic social context.	To train typically developing children to implement reciprocal imitation training with their siblings with autism spectrum disorder.	15-to-30-minute session per week, for 10 weeks at home.	The trainer used a manual written in child-friendly language and augmented with pictures depicting the intervention techniques. The trainer explained the technique and read through the relevant portion of	Not listed.	Based on literature.	The trainer (first author) was a graduate-level student with about 2 years of experience working with young children with autism spectrum disorder and	Yes

				the manual with the TD sibling, role-played the technique with the TD sibling while giving instruction and feedback, and demonstrated the technique with the sibling with autism spectrum disorder while explaining the actions. After each technique was taught, a poster depicting the technique was placed in the playroom to remind the TD sibling to use the technique. Feedback was provided to the TD sibling at certain periods.			trained in the intervention.	
Lewandowski et al. 2014 [87] Comic Strip Conversation Intervention	To address sibling conflict and support Theory of Mind development through learning about the social and emotional factors that contribute to both conflict and resolution.	Same objective as the sibling with a neurodisability.	12 times for each phase, two phases in total at home.	The researcher introduced and explained the symbols dictionary in the first few sessions to remind the TD sibling and sibling with a neurodisability of the talking and thinking bubbles that would be used in the session. After the participants	Not listed.	Based on comic strip conversation intervention described in the literature.	Researcher (first author).	No

				showed understanding of the established conventions, the symbols dictionary was left on the table but was not reviewed unless the participants had questions. The activity was introduced by explaining that the researcher would help the TD sibling and sibling with a neurodisability to write and draw while discussing a challenging situation. Both the TD sibling and sibling with a neurodisability explained the events that took place during the challenging situation, and participated in the writing and drawing as much as possible.				
Özen 2015 [109]	Target skills were taught such as how to perform a behaviour independently,	To teach social interaction skills, including using effective prompts, taking turns, or	29 sessions at home.	Activities including watching sample video clips to teach social interaction skills, having the researcher describe	Not listed.	Not listed.	Researcher with a background of special education, and has 23 years of	Yes

	taking turns, or saying appropriate phrases.	reinforcing appropriate play behaviours with the sibling with autism spectrum disorder.		the social interaction behaviour, providing opportunities for the TD siblings to ask questions, and having the TD sibling and the sibling with autism spectrum disorder play together on the iPad. The researcher provided feedback on the behaviours from the TD sibling and sibling with autism spectrum disorder.			work experience with children with autism spectrum disorder and their families.	
Kryzak et al. 2017 [86] Behavioral Skills Training	To improve reciprocal interactions between siblings.	To improve self-management (goal setting, monitoring, and recruiting reinforcement) of the Stay-Play-Talk curriculum.	Weekly sessions for 14 weeks at home.	The researcher reviewed the self-management task analysis with the TD sibling along with that day's lesson. After introducing the lesson, the researcher modeled the responses described in the curriculum. The TD sibling then rehearsed the lesson with feedback from the researcher. Then, the TD sibling proceeded to play games with	Not listed.	Modified curriculum described in the literature.	First author with a background in psychology and forensic sciences, and experience with applied behaviour analysis.	No

				their sibling with autism spectrum disorder.				
Neff et al. 2017 [107]	To learn on-task behavior.	To train TD siblings to provide positive reinforcement and prompts during play sessions with their sibling with autism spectrum disorder.	3-5 min videos that are watched prior to 15-minute sessions at home and treatment center.	Customized videos were made for each pair of TD sibling and sibling with autism spectrum disorder. Each video consisted of at least three examples of physical prompting, gestural prompting, modeling prompting, and verbal prompting demonstrated by the researcher in the video. There were at least six examples of positive reinforcement modeled by the researcher across prompted and independent responses. The TD sibling watched the video corresponding to each game or activity immediately before	Not listed.	Not listed.	Researcher	No

				playing the game with their sibling with autism spectrum disorder. Additional teaching was comprised of practice sessions with the researcher and feedback.				
Akers et al. 2018 [19]	To increase the number of appropriate vocalizations emitted by siblings with autism spectrum disorder.	To implement the script-fading procedure with their sibling with autism spectrum disorder.	Three sessions per day at home. Training ended when the sibling correctly implemented each component with their parent with 95% or better accuracy. The siblings met mastery after one session that took approximately 30 minutes.	The TD siblings were trained to: (a) orient to the sibling with autism spectrum disorder during play, (b) refrain from asking questions or giving directions, (c) respond to all of the verbalizations from their sibling with autism spectrum disorder, and (d) to comment on their own play actions. Siblings were instructed to present an auditory script and provide verbal prompts for their sibling with autism spectrum disorder to respond to.	Not listed.	Not listed.	Researcher. Parents of the siblings served as a research assistant.	No

Douglas et al. 2018 [46]	To increase sibling talk.	To increase sibling talk, as well as the frequency/percentage of responses to the sibling with complex communication needs.	One 45-min session and one 30-min session for a total of two training sessions, scheduled two days apart, conducted at home.	The first session provided a description of the strategy steps, visual text on a PowerPoint, video demonstration, and a handout of the training content. Verbal practice, questions, and application activities were included for each strategy step. The second session included a quiz of the strategy steps, role-play of the strategy during a play activity with the trainer, and a practice play session with feedback with their sibling with complex communication needs.	Not listed.	Based on the literature and adaptation of other sibling programs.	First author.	Yes
Spector et al. 2018 [127]	To increase imitative utterances, vocalizations, and speech.	To implement Natural Learning Paradigm using a video.	20–25-minute training session; 5 minute sessions of sibling-mediated intervention for a total	Siblings were trained to implement Natural Learning Paradigm using a video that role-modeled the strategies. Each sibling was told they will be taught to play a game with	Not listed.	Based on the literature and adaptation of other sibling programs.	Adult therapist.	No

			of 20 sessions, at an after school behavioural management center.	their sibling with autism spectrum disorder. The sibling was tested for comprehension by role-playing the procedures in the video. Siblings were then asked to play a game with their sibling with autism spectrum disorder using the strategies from the video.				
Daffner et al. 2020 [40]	To increase the frequency of positive social behaviours and decrease the frequency of negative social behaviours.	To teach specific behaviour strategies and prompting skills for facilitating social interactions with the child with attention deficit hyperactivity disorder.	15-30 minutes sessions twice a week for approximately 17-22 sessions in total at home.	Siblings were taught three lessons focused on specific social behavior strategies to help their brother or sister with attention deficit hyperactivity disorder in play or social interactions, such as sharing, giving or asking for help, and compromising. The researcher explained the strategy, used videos to demonstrate the strategy, and allowed the TD sibling to practice the strategy with feedback from the	Not listed.	Modification of peer-mediated intervention programs from the literature.	First author	Yes

				researcher. The TD sibling then played with their sibling with autism spectrum disorder after the training with no additional feedback.				
Tsao 2020 [136]	To increase their social interactions with their TD siblings.	To learn and implement social skills strategies while playing with their siblings with developmental disabilities.	15-20 minutes for 5-7 sessions in total at home.	The first two lessons introduced the three steps of the curriculum, and the following lessons focused on specific strategies to involve the siblings with developmental disabilities such as offering to help and requesting assistance. The researcher reviewed the previous lesson, introduced the skill/strategy, and read a story illustrating the use of the skill/strategy. After the training sessions, the researcher encouraged the TD siblings to use the discussed strategies when playing with their siblings with developmental disabilities.	Not listed.	Based on literature.	Researcher.	No

Table 5. Outcomes of programs focused on empowerment, and key findings for the typically developing (TD) siblings.

Study	Program Outcomes	Key findings for the TD siblings	Key findings for the siblings with a neurodisability
Weinrott 1974 [138]	Knowledge acquisition, skill development, satisfaction with the program, empowerment.	Moderate or vast improvement in the quality of their interaction with their sibling with an intellectual disability. In nearly two-thirds of the families, siblings were observed to be spending more time with their sibling with an intellectual disability. All siblings, with one exception, placed demands upon their sibling with an intellectual disability to use newly acquired speech, to identify concepts, and to follow directions. Siblings exhibited more patience than before. Four siblings began serving as aides in their sibling with an intellectual disability in the classroom at school. Another two siblings selected special education as their prospective major in college, which was not made before the program and for one of the sibling, it was not considered.	Not listed.
Doleys et al. 1975 [45]	Implementation of a program by the sibling without a neurodisability	This program could be designed and successfully carried out by the TD sibling in a natural environment.	The verbal behaviour of an adolescent with intellectual disability could be modified with the technique of response cost contingency.
Miller et al. 1976 [106]	Skill development	Decreased family arguing and increased positive interactions between siblings.	Able to play alone for as long as 45 minutes., was more outgoing and assertive in family interactions. Positive effects were maintained 6-months after.
Colletti et al. 1977 [36]	Skill development	Siblings can modify the behaviour of their siblings with a neurodisability. In Experiment 1, a sibling delivered contingent reinforcement which dramatically increased the ability to carry out the task (i.e., bead stringing by the sister with autism spectrum disorder). In Experiment 2, two siblings were able to work effectively with their brother with a neurodisability on separate tasks.	Not listed.

Schreibman et al. 1983 [124]	Skill development	The siblings became proficient in behavioral teaching skills.	There were improvements in the correct responding. Skills could be applied in different environments, and in a much less structured type of interaction than during the training sessions.
Lobato et al. 1985 [95]	Skill development	The TD sibling was effective in teaching the sibling with a neurodisability basic self-care skills.	The results of the second intervention on bedmaking were indicating a positive trend but were prematurely terminated due to a separate injury. No issues with noncompliance during structured training sessions with the TD sibling.
James et al. 1986 [75]	Skill development	The TD siblings easily acquired the requisite skills with increased initiations for interactions.	The baseline data for the handicapped siblings showed that their major deficit was initiations rather than responsiveness. Although the siblings with disabilities rarely initiated interactions with their TD siblings, they did respond appropriately to initiations made by their TD siblings. The interactions between siblings generalized to larger play groups and across siblings.
Swenson-Pierce et al. 1987 [131]	Skill development	Successful training to use increased prompting and social praise with a high degree of accuracy. Enjoyment in participating in the study, which did not interfere significantly with personal time. Two TD siblings indicated that they used their skills when assisting their sibling with a neurodisability with other tasks.	The siblings were able to perform the skills more independently.
Clark et al. 1989 [33]	Skill development, sibling attitude	Increased use of attending strategies and sign languages, and decreased use of controlling strategies. There was variation across sibling pairs with respect to the level of maintenance of these behaviors at follow-up.	Reductions in behavioral problem severity. Parents reported increases in positive interactions between siblings, which was maintained at six-months.

Craft et al. 1990 [37]	Knowledge acquisition and skill development	Increased self-confidence.	Increased family interactional patterns through self-reports and family functioning from parent reports. Indirect benefits of improved physical functioning. Reports that the TD siblings understood them better, spent more time with them, and were more patient with them. Some parents felt that siblings gained an increased sense of importance and came to know how much the sibling interactions meant to their family.
Coe et al. 1991 [35]	Skill development	TD siblings quickly mastered both prompt and reinforcement techniques and were able to employ them without direct adult supervision.	Low frequency of verbal play behaviour and variable nonverbal play. By the end of the study, nonverbal and verbal play responses occurred in over 72% of recording intervals.
Celiberti et al. 1993 [30]	Skill development	Demonstrated rapidly mastery of target skills and sustained maintenance of skills as training shifted to other targeted areas and during follow-up probes, and able to apply skills to other types of interactions with the sibling with a neurodisability (e.g., at dinnertime, outdoor activities). Became more comfortable interacting with sibling with autism spectrum disorder.	Not applicable.
Hancock et al. 1996 [69]	Skill development	All TD siblings learned the milieu language teaching techniques and implemented these procedures in a play setting with their siblings with a neurodisability. They all reported that they enjoyed spending time with their siblings with a neurodisability in a play situation and that they learned about their siblings' competencies from the experience of being involved in the intervention.	When the teaching behaviors by the TD sibling, teaching continues to affect the language behaviors demonstrated by the siblings with a neurodisability.

Trent et al. 2005 [132]	Skill development	TD siblings learned the responsive interaction techniques quickly and used them in interactions with their younger siblings with Down syndrome during intervention. TD siblings increased their use of mirroring and verbal responding during play interactions. When training for verbal responding was introduced to the target siblings, the number of intervals during which they used mirroring decreased. Responsive interaction strategies were maintained at 1-month follow-up.	Modest effects on their verbal behaviors and maintained at 1-month follow-up.
Tsao et al. 2006 [134]	Skill development	Moderate support for the effectiveness of a social intervention that involved TD siblings as mediators of social interactions for the siblings with autism spectrum disorder. The program did encourage more social interactions between TD siblings and their siblings with autism spectrum disorder. For three of the four siblings, social initiations toward their siblings with autism spectrum disorder increased during the program intervention phase.	Modest, positive changes in the social interactions of three siblings with autism spectrum disorder. Modest evidence for maintenance of social behavior. Clear increases in joint attention for three siblings. Limited evidence of social behaviour responses in other settings.
Stewart et al. 2007 [129]	Skill development	The TD sibling was able to prompt appropriate conversational behavior in the natural environment.	Appropriate eye contact was made during 78.6% of intervals, asked whether the partner was bored during 53.6% of intervals, asked whether the student assistant wished to change the topic during 53.6% of intervals, and always avoided perseverative topics.

Trent-Stainbrook et al. 2007 [133]	Skill development	Responsive interaction techniques were quickly learned and used them in interactions with the siblings with a neurodisability. Use of these strategies were maintained at 1-month follow-up for all TD siblings. The effects of the intervention in the play setting did not readily generalize to the snack setting. Sibling interactions during generalization sessions were positive, but the siblings took very asymmetrical roles. The younger siblings often needed assistance preparing their own snacks. The older TD siblings often assumed the role of a teacher or helper, instructing the younger siblings on how to prepare the snack rather than being responsive to their acts of intentional communication.	Slight increase in the number of comments made in each session. No change occurred in the number of requests made by the younger siblings with Down syndrome. The quality of interactions between siblings appeared to improve.
Tsao et al. 2010 [135]	Skill development	Not applicable.	Not applicable.
Ferraioli et al. 2011 [53]	Skill development	TD siblings implemented the intervention with high fidelity for most components, although it was difficult for them to remember all components without prompts (e.g., providing differential praise). Siblings were generally able to understand and apply concepts of obtaining and maintaining attention, providing tangible reinforcement, and persisting. All the siblings found the treatment acceptable by indicating during interviews that teaching was fun and that they would continue using the skills after the conclusion of the study.	There was meaningful change in the responding skills, with maintenance of acquired skills for responding for all siblings with a neurodisability and initiating from some siblings with a neurodisability. Performance reflected individual variation and response.
Chu et al. 2012 [31]	Skill development	Enhanced aquatic skills.	Increased physical and social interactions.

Oppenheim-Leaf et al. 2012 [108]	Skill development	Ability to demonstrate all skills taught with a teacher and the sibling with autism spectrum disorder. Play interactions between the TD sibling and sibling with autism spectrum disorder increased during free-play situations. Generalization of training to the free-play situation varied between siblings.	Performance of the siblings with autism spectrum disorder during generalization probes suggested that the TD siblings were effective at encouraging their siblings with autism spectrum disorder to engage in targeted social behaviours. Following the program, the siblings with autism spectrum disorder frequently engaged in the targeted social behavior, either independently or following a prompt from the TD sibling.
Walton et al. 2012 [137]	Skill development	Varying success learning to use the three intervention strategies independently. Reported enjoyment of the intervention, although the skills were a little hard to learn.	Some siblings with autism spectrum disorder showed increases in certain behaviours. None of the siblings with autism spectrum disorder maintained their skill gains when interacting with a different child. However, many of the sibling's gains were maintained (or even continued to increase) at the 1-month follow-up.
Lewandowski et al. 2014 [87]	Knowledge acquisition	No effect of intervention.	No effect of intervention. During the co-occurring intervention for both siblings, the sibling with a neurodisability showed improvements in early and basic Theory of Mind competencies, which is the ability to read and interpret thoughts and feelings of self and others. According to parent report, the sibling with neurodisability also demonstrated the ability to better able negotiate challenging situations with his TD sibling.

Özen 2015 [109]	Skill development	Demonstrated ability to perform social interaction skills with 85-91% accuracy, which were maintained 1-week and 2-weeks after the program. Skills were generalizable to different children with autism spectrum disorder.	Siblings with autism spectrum disorder demonstrated varying abilities to acquire skills, including following directions, taking turns, responding appropriately to learning opportunities. Targeted skills were maintained 1-week and 2-week after the program.
Kryzak et al. 2017 [86]	Skill development	Improved self-management (goal setting, monitoring, and recruiting reinforcement) of a social skills curriculum. Improvements were largely maintained and generalized to different settings.	Some corresponding improvements in reciprocal interaction of both siblings. Siblings with autism spectrum disorder also showed improved reciprocal interactions after intervention that were largely maintained through the 14-week maintenance sessions.
Neff et al. 2017 [107]	Skill development	Video modeling alone served as an effective teaching device for teaching prompting and reinforcement skills during play for two of the three TD siblings. Learned skills were generalizable to novel games.	When the TD siblings increased their use of appropriate prompts and reinforcement, the task behavior of their sibling with autism spectrum disorder subsequently increased. When supplemental reinforcement was introduced for the TD sibling, the delivery of appropriate prompts and reinforcement from the TD sibling increased, the task behavior of the sibling with autism spectrum disorder also increased.
Akers et al. 2018 [19]	Knowledge acquisition and maintenance of	Demonstrated ability to implement procedures with fidelity.	The number of contextually appropriate statements increased.

	script fading procedures for the sibling with autism spectrum disorder at follow-up. Skill development.		
Douglas et al. 2018 [46]	Skill development	Increased frequency of sibling communication supports.	Increased communication for the siblings with complex communication needs.
Spector et al. 2018 [127]	Skill development	Demonstrated ability to effectively learn the training techniques from the Natural Learning Paradigm in a short amount of time and engage in these techniques with their sibling with autism spectrum disorder.	Two of three children with autism spectrum disorder showed increases in speech and verbalization. Generalization of treatment gains was limited.
Daffner et al. 2020 [40]	Satisfaction with the program from the perspectives of the parents and sibling participants. Skill development for the siblings.	All three TD siblings either agreed or strongly agreed that the strategies they learned to use were fair, the strategies helped them and the sibling with attention deficit hyperactivity disorder be better playmates, and learning to use the strategies was easy. Increased use of several positive social behaviours including sharing, asking/giving help, and compromising. Siblings were observed to successfully learn and use these specific social skill strategies with relatively high integrity.	Promoted positive social behaviours and social skills.
Tsao 2020 [136]	Skill development	Moderate support for the effectiveness of a social interaction intervention that involved TD siblings as mediators of social interactions for children with developmental disabilities. Medium effect on the application of strategies by TD siblings. The intervention did encourage more social interactions between typically developing brothers and children with developmental disabilities.	Some effect on the social behaviors of children with DD. There was a small effect on social behaviors of children with developmental disabilities and their TD siblings.

REFERENCES

1. UNICEF. The state of the world's children 2006: excluded and invisible. New York: United Nations Children's Fund; 2005.
2. Morris C, Janssens A, Tomlinson R, Williams J, Logan S. Towards a definition of neurodisability: a Delphi survey. Dev Med Child Neurol. 2013;55:1103–8.
3. Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. Am Psychol. 2000;55:469–80.
4. Freeman M, Stewart D, Cunningham CE, Gorter JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. J Transit Med. 2018;1.
5. Rosenbaum P, King S, Law M, King G, Evans J. Family-centred service. Phys Occup Ther Pediatr. Taylor & Francis; 1998;18:1–20.
6. Bowen M. Clinical view of the family. Fam Syst. 1995;2:153–6.
7. Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. Intellect Dev Disabil. 2009;47:208–19.
8. Davys D, Haigh C. Older parents of people who have a learning disability: perceptions of future accommodation needs. Br J Learn Disabil. 2008;36:66–72.
9. Redquest BK, Tint A, Ries H, Goll E, Rossi B, Lunsky Y. Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. J Policy Pract Intellect Disabil. 2020;17:239–46.
10. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. J Appl Res Intellect Disabil. 2018;31:423–34.
11. D'Arcy F, Flynn J, McCarthy Y, O'Connor C, Tierney E. Sibshops. An evaluation of an interagency model. J Intellect Disabil. 2005;9:43–57.
12. Conroy-Harris A. England and Wales. Adopt Foster. Sage Publications, Ltd.; 2019;43:91–4.
13. Lobato DJ, Kao BT. Integrated sibling-parent group intervention to improve sibling knowledge and adjustment to chronic illness and disability. J Pediatr Psychol. 2002;27:711–6.
14. Sheikh R, Patino V, Cengher M, Fiani T, Jones EA. Augmenting sibling support with parent-sibling training in families of children with autism. Dev Neurorehabil. 2019;22:542–52.
15. Peters M, Godfrey C, McInerney P, Munn Z, Tricco A, Khalil H. Scoping Reviews. In: Aromataris E, Munn Z, editors. Joanna Briggs Inst Rev Man. JBI Manual for Evidence Synthesis; 2020.
16. Nguyen L, Bootsma J, Rezze B Di, Jack S, Ketelaar M, Gorter JW. Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: Protocol of a scoping review. BMJ Open. 2021;11:e053184.
17. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA extension for scoping reviews (PRISMA-ScR): Checklist and explanation. Ann Intern Med. 2018;169:467–73.
18. Canadian Institutes of Health Research. Guide to knowledge translation planning at CIHR: Integrated and end-of -grant approaches [Internet]. 2015 [cited 2019 Apr 22]. Available from: http://www.cihr-irsc.gc.ca/e/45321.html#a3
19. Akers JS, Higbee TS, Pollard JS, Reinert KS. Sibling-implemented script fading to promote play-based statements of children with autism. Behav Anal Pract. 2018;11:395–9.
20. Brouzos A, Vassilopoulos SP, Tassi C. A psychoeducational group intervention for siblings of children with autism spectrum disorder. J Spec Gr Work. 2017;42:274–98.
21. Ross P, Cuskelly M. Adjustment, sibling problems and coping strategies of brothers and

sisters of children with autistic spectrum disorder. J Intellect Dev Disabil. 2006;31:77–86.
22. Perry A. What it's like to have a brother or sister with a developmental disorder. Toronto, Ontario, Canada;
23. Goodman R, Meltzer H, Bailey V. The strengths and difficulties questionnaire: A pilot study on the validity of the self-report version. Eur Child Adolesc Psychiatry. 1998;7:125–30.
24. Burke MM, Lee CE, Carlson SR, Arnold CK. Exploring the preliminary outcomes of a sibling leadership program for adult siblings of individuals with intellectual and developmental disabilities. Int J Dev Disabil. 2018;0:1–8.
25. Burke M, Arnold C, Owen A. Identifying the correlates and barriers of future planning among parents of individuals with intellectual and developmental disabilities. Intellect Dev Disabil. 2018;56:90–100.
26. Koren PE, DeChillo N, Friesen BJ. Measuring empowerment in families whose children have emotional disabilities: A brief questionnaire. Rehabil Psychol. 1992;37:305–21.
27. Burke MM, Fish T, Lawton K. A comparative analysis of adult siblings' perceptions toward caregiving. Intellect Dev Disabil. 2015;53:143–57.
28. Perry JL. Measuring public service motivation: An assessment of construct reliability and validity. J Public Adm Res Theory. 1996;6:5–22.
29. Taylor JL, Hodapp RM, Burke M, Rabideau C. Ecological approaches to transition intervention in ASD: Training parents to more effectively navigate the service system. [Internet]. 2016 [cited 2021 Dec 7]. Available from: https://insar.confex.com/insar/2016/videogateway.cgi/id/799?recordingid=799
30. Celiberti DA, Harris SL. Behavioral intervention for siblings of children with autism: A focus on skills to enhance play. Behav Ther. 1993;24:573–99.
31. Chu CH, Pan CY. The effect of peer- and sibling-assisted aquatic program on interaction behaviors and aquatic skills of children with autism spectrum disorders and their peers/siblings. Res Autism Spectr Disord. 2012;6:1211–23.
32. Humphries KM. Huphries' assessment of aquatic readiness. Texas Women's University; 2008.
33. Clark ML, Cunningham LJ, Cunningham CE. Improving the social behavior of siblings of autistic children using a group problem solving approach. Child Fam Behav Ther. 1989;11:19–33.
34. Rosenbaum PL, Armstrong RW, King SM. Children's attitudes toward disabled peers: A self-report measure. J Pediatr Psychol. 1986;11:517–30.
35. Coe DA, Matson JL, Craigie CJ, Gossen MA. Play skills of autistic children: Assessment and instruction. Child Fam Behav Ther. 1991;13:13–40.
36. Colletti G, Harris SL. Behavior modification in the home: Siblings as behavior modifiers, parents as observers. J Abnorm Child Psychol. 1977;5:21–30.
37. Craft MJ, Lakin JA, Oppliger RA, Clancy GM, Vanderlinden DW. Siblings as change agents for promoting the functional status of children with cerebral palsy. Dev Med Child Neurol. 1990;32:1049–57.
38. Crouthamel CS. Siblings of handicapped children: A group support program. Early Child Dev Care. 1988;37:119–31.
39. Piers VE, Harris DB. Children's Self-Concept Scale: Revised Manual. Los Angeles: Western Psychological Services; 1984.
40. Daffner MS, DuPaul GJ, Kern L, Cole CL, Cleminshaw CL. Enhancing social skills of young children with ADHD: Effects of a sibling-mediated intervention. Behav Modif.

2020;44:698–726.
41. Connors CK. Connors early childhood manual. Toronto: Multi-Health Systems; 2009.
42. DuPaul GJ, Power TJ, Anastopoulos AD, Reid R. ADHD rating scale-5 for children and adolescents: Checklists, norms, and clinical interpretation. New York: Guilford; 2016.
43. Rutter M, Bailey A, Lord C. Social communication questionnaire (SCQ). Los Angeles: Western Psychological Services; 2003.
44. Barkley RA, Murphy KR. Attention-deficit hyperactivity disorder: A clinical workbook. 3rd ed. New York: Guilford; 2006.
45. Doleys DM, Slapion MJ. The reduction of verbal repetitions by response cost controlled by a sibling. J Behav Ther Exp Psychiatry. 1975;6:61–3.
46. Douglas SN, Kammes R, Nordquist E, D'Agostino S. A pilot study to teach siblings to support children with complex communication needs. Commun Disord Q. 2018;39:346–55.
47. Sigafoos J, Arthur-Kelly M, Butterfield N. Enhancing everyday communication for children with disabilities. Baltimore, MD: Brookes; 2006.
48. Furman W, Buhrmester D. Children's perceptions of the qualities of sibling relationships. Child Dev. 1985;56:448.
49. Dyson LL. A support program for siblings of children with disabilities: What siblings learn and what they like. Psychol Sch. 1998;35:57–65.
50. Evans J, Jones J, Mansell I. Supporting siblings: Evaluation of support groups for brothers and sisters of children with learning disabilities and challenging behaviour. J Learn Disabil. 2001;5:69–78.
51. Bene E, Anthony J. Family Relations Test: Children's Version. Windsor: NFER Nelson;
52. Battle J. Culture Free Self Esteem Inventories for Children and Adults. Seattle and Windsor: NFER Nelson; 1981.
53. Ferraioli SJ, Harris SL. Teaching joint attention to children with autism through a sibling-mediated behavioral intervention. Behav Interv. 2011;26:261–81.
54. Mundy P, Sigman M, Ungerer J, Sherman T. Defining the social deficits of autism: The contribution of non-verbal communication measures. J Child Psychol Psychiatry. 1986;27:657–69.
55. Fjermestad K, Pat P, Dearozet S, Vatne T, Hafting M, Jegannathan B. Manual-Based Group Intervention for Siblings and Parents of Children with Neurodevelopmental Disorders in Cambodia. J Dev Phys Disabil. 2021;33:839–56.
56. Goodman R. Psychometric Properties of the Strengths and Difficulties Questionnaire. J Am Acad Child Adolesc Psychiatry. 2001;40:1337–45.
57. Lovibond PF, Lovibond SH. The structure of negative emotional states: Comparison of the Depression Anxiety Stress Scales (DASS) with the Beck Depression and Anxiety Inventories. Behav Res Ther. Pergamon; 1995;33:335–43.
58. Loeber, R., Farrington, D. P., Stouthamer-Loeber, M., & Van Kammen WB. Antisocial behavior and mental health problems: Explanatory factors in childhood and adolescence. Psychology Press; 1998.
59. Fjermestad KW, Silverman WK, Vatne TM. Group intervention for siblings and parents of children with chronic disorders (SIBS-RCT): Study protocol for a randomized controlled trial. Trials. 2020;21:1–12.
60. Ravens-Sieberer U, Bullinger M. Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. Qual Life Res. 1998;7:399–407.

61. Sahler OJ, Roghmann KJ, Carpenter PJ, Mulhern RK, Dolgin MJ, Sargent JR, et al. Sibling adaptation to childhood cancer collaborative study: prevalence of sibling distress and definition of adaptation levels. J Dev Behav Pediatr. 1994;15:353–66.
62. Gettings S, Franco F, Santosh PJ. Facilitating support groups for siblings of children with neurodevelopmental disorders using audio-conferencing: A longitudinal feasibility study. Child Adolesc Psychiatry Ment Health. 2015;9(1):1-15.
63. Gringras P, Santosh P, Baird G. Development of an Internet-based real-time system for monitoring pharmacological interventions in children with neurodevelopmental and neuropsychiatric disorders. Child Care Health Dev. 2006;32:591–600.
64. Goodman A, Goodman R. Strengths and difficulties questionnaire as a dimensional measure of child mental health. J Am Acad Child Adolesc Psychiatry. Elsevier; 2009;48:400–3.
65. Varni JW, Seid M, Kurtin PS. PedsQL^{TM} 4.0: Reliability and validity of the Pediatric Quality of Life Inventory^{TM} version 4.0 generic core scales in healthy and patient populations. 2001.
66. Granat T, Nordgren I, Rein G, Sonnander K. Group intervention for siblings of children with disabilities: a pilot study in a clinical setting. Disabil Rehabil. 2012;34:69–75.
67. Lobato DJ, Kao BT. Brief report: Family-based group intervention for young siblings of children with chronic illness and developmental disability. J Pediatr Psychol. 2005;30:678–82.
68. Buhrmester D, Furman W. Perceptions of sibling relationships during middle childhood and adolescence. Child Dev. 1990;61:1387–98.
69. Hancock TB, Kaiser AP. Siblings' use of milieu teaching at home. Topics Early Child Spec Educ. 1996;16:168–90.
70. Hayden NK, McCaffrey M, Fraser-Lim C, Hastings RP. Supporting siblings of children with a special educational need or disability: An evaluation of Sibs Talk, a one-to-one intervention delivered by staff in mainstream schools. Support Learn. 2019;34:404–20.
71. Goodman R. The Strengths and Difficulties Questionnaire: A research note. J Child Psychol Psychiatry. John Wiley & Sons, Ltd; 1997;38:581–6.
72. Goodman R. The extended version of the strengths and difficulties questionnaire as a guide to child psychiatric caseness and consequent burden. J Child Psychol Psychiatry. 1999;40:791–9.
73. Allen K, Marlow R, Edwards V, Parker C, Rodgers L, Ukoumunne OC, et al. "How I Feel About My School": The construction and validation of a measure of wellbeing at school for primary school children. Clin Child Psychol Psychiatry. SAGE PublicationsSage UK: London, England; 2018;23:25–41.
74. Ford T. "How I Feel About My School" (HIFAMS) questionnaire [Internet]. 2013 [cited 2021 Dec 7]. Available from: http://medicine.exeter.ac.uk/hifams/
75. James SD, Egel AL. A direct prompting strategy for increasing reciprocal interactions between handicapped and nonhandicapped siblings. J Appl Behav Anal. 1986;19:173–86.
76. Jones EA, Fiani T, Stewart JL, Neil N, McHugh S, Fienup DM. Randomized controlled trial of a sibling support group: Mental health outcomes for siblings of children with autism. Autism. 2020;24:1468–81.
77. Kovacs M. The children's depression inventory. 2nd ed. Western Psychological Services; 2011.
78. Reynolds CR, Richmond BO. Revised children's manifest anxiety scale. 2nd ed. Pro-Ed; 2008.
79. Ayers TS, Sandier IN, West SG, Roosa MW. A dispositional and situational assessment of children's coping: Testing alternative models of coping. J Pers. 1996;64:923–58.
80. Achenbach, T.M. Rescorla LA. Manual for the ASEBA preschool forms & profiles: An

integrated system of multi-informant assessment. University of Vermont; 2000.
81. Achenbach, T.M. Rescorla LA. Manual for the ASEBA school-age forms & profiles: An integrated system of multi-informant assessment. University of Vermont; 2001.
82. Schopler E, Van Bourgondien ME. Childhood Autism Rating Scale(TM). 2nd ed. Western Psychological Services; 2010.
83. Kryzak LA, Cengher M, Feeley KM, Fienup DM, Jones EA. A community support program for children with autism and their typically developing siblings: Initial investigation. J Intellect Disabil. 2015;19:159–77.
84. Kovacs M. The children's depression inventory. Psychopharmacology (Berl). 1985;21:995–8.
85. Glasberg BA. The Development of Siblings' Understanding of Autism Spectrum Disorders. J Autism Dev Disord. Kluwer Academic Publishers-Plenum Publishers; 2000;30:143–56.
86. Kryzak LA, Jones EA. Sibling self-management: Programming for generalization to improve interactions between typically developing siblings and children with autism spectrum disorders. Dev Neurorehabil. 2017;20:525–37.
87. Lewandowski JF, Hutchins TL, Prelock PA, Murray-Close D. Examining the benefit of including a sibling in story-based intervention with a child with Asperger Syndrome. Contemp Issues Commun Sci Disord. 2014;41:179–95.
88. Gilliam JE. Gilliam Autism Rating Scale. 2nd ed. Austin, TX: Pro-Ed; 2006.
89. Constantino JN, Gruber CP. The social responsiveness scale. Los Angeles, CA: Western Psychological Services; 2005.
90. Gresham FM, Elliott SN. Social skills rating system. Circle Pines, MN: AGS; 1990.
91. Hutchins TL, Prelock PA, Bonazinga L. Psychometric Evaluation of the Theory of Mind Inventory (ToMI): A Study of Typically Developing Children and Children with Autism Spectrum Disorder. J Autism Dev Disord. 2012;42:327–41.
92. Williams KT. Expressive vocabulary test. Minneapolis, MN: NCS Pearson; 2007.
93. Dunn LM, Dunn D. Peabody picture vocabulary test. 4th ed. Minneapolis, MN: NCS Pearson; 2007.
94. Bishop D. Test for reception of grammar. 2nd ed. London, UK: The Psychological Corporation; 2003.
95. Lobato D, Tlaker A. Sibling intervention with a retarded child. Educ Treat Child. 1985;8:221–8.
96. Lobato D. Brief report: Preschool siblings of handicapped children - Impact of peer support and training. J Autism Dev Disord. 1985;15:345–50.
97. Wijma CA, Masino LL, Hodapp RM. Families of children with 5p- (cri du chat) syndrome: familial stress and sibling reactions. Dev Med Child Neurol. 2008;39:757–61.
98. Achenbach TM. Manual for the Child Behavior Checklist/4-18 and 1991 Profile. Burlington: University of Vermont; 1991.
99. Harter S, Pike R. The pictorial scale of perceived competence and acceptance for young children. Denver, CO: University of Denver; 1983.
100. McCullough K, Simon SR. Feeling heard: a support group for siblings of children with developmental disabilities. Soc Work Groups. 2011;34:320–9.
101. McLinden SE, Miller LM, Deprey JM. Effects of a support group for siblings of children with special needs. Psychol Sch. 1991;28:230–7.
102. Piers EV, Harris DE. The Piers-Harris Children's Self-Concept Scale (The Way I Feel About Myself). Los Angeles: Western Psychological Services; 1969.

103. Hodges B, Mott DW, Jenkins V. Let's Grow Together. 1982.
104. McLinden SE. Who Helps Me. 1987.
105. Achenbach TM, Edelbrock CS. Manual for the Child Behavior Checklist and Revised Child Behavior Profile. Burlington: VT; 1983.
106. Miller NB, Cantwell DP. Siblings as therapists: a behavioral approach. Am J Psychiatry. United States; 1976;133:447–50.
107. Neff ER, Betz AM, Saini V, Henry E. Using video modeling to teach siblings of children with autism how to prompt and reinforce appropriate play. Behav Interv. 2017;32:193–205.
108. Oppenheim-Leaf ML, Leaf JB, Dozier C, Sheldon JB, Sherman JA. Teaching typically developing children to promote social play with their siblings with autism. Res Autism Spectr Disord. Elsevier Ltd; 2012;6:777–91.
109. Özen A. Effectiveness of siblings-delivered ipad game activities in teaching social interaction skills to children with autism spectrum disorders. Educ Sci Theory Pract. 2015;15:1287–303.
110. Phillips RSC. Intervention with siblings of children with developmental disabilities from economically disadvantaged families. Fam Soc. 1999;80:569–77.
111. Dubois DL, Felner RD, Brandy S, Phillips RSC, Lease AM. Early adolescent self-esteem: A developmental-ecological framework and assessment strategy. J. Res. Adolesc. 1996.
112. Procidino, M. E., & Heller K. Measures of perceived social support from friends and from family: Three validation studies. Am J Community Psychol. 1983;11:1–24.
113. Rowlison, R. T., & Felner RD. Major life events, hassles, and adaptation in adolescence: Confounding in the conceptualization and measurement of life stress and adjustment revisited. J Pers Soc Psychol. 1988;55:432–44.
114. Moos RH, Moos BA. The Family Environment Scale manual. 2nd ed. Palo Alto: Consulting Psychologist Press; 1986.
115. Roberts RM, Ejova A, Giallo R, Strohm K, Lillie M, Fuss B. A controlled trial of the SibworkS group program for siblings of children with special needs. Res Dev Disabil. Pergamon Press - An Imprint of Elsevier Science; 2015;43:21–31.
116. Harter S. The self-perception profile for children: Revision of the perceived competence scale for children. Denver, CO: University of Denver; 1985.
117. Causey DL, Dubow EF. Development of a self-report coping measure for elementary school children. J Clin Child Psychol. Lawrence Erlbaum Associates, Inc.; 1992;21:47–59.
118. Rosenberg M. Society and the adolescent self-image. Reprint. Middletown, CT: Wesleyan University Press; 1989.
119. Taffe JR, Gray KM, Einfeld SL, Dekker MC, Koot HM, Emerson E, et al. Short form of the Developmental Behaviour Checklist. Am J Ment Retard. 2007;112:31–9.
120. Australian Bureau of Statistics. Socio-economic Indexes for Areas (SEIFA). 2006.
121. Marsland AL, Long KA, Howe C, Thompson AL, Tersak J, Ewing LJ. A pilot trial of a stress management intervention for primary caregivers of children newly diagnosed with cancer: Preliminary evidence that perceived social support moderates the psychosocial benefit of intervention. J Pediatr Psychol. Oxford Academic; 2013;38:449–61.
122. Roberts RM, Ejova A, Giallo R, Strohm K, Lillie ME. Support group programme for siblings of children with special needs: predictors of improved emotional and behavioural functioning. Disabil Rehabil. 2016;38:2063–72.
123. Rye K, Hicks S, Falconer C. Evaluating a group for young people who have a sibling with a disability. Learn Disabil Pract. 2018;1.

124. Schreibman L, O'Neill RE, Koegel RL. Behavioral training for siblings of autistic children. J Appl Behav Anal. United States; 1983;16:129–38.
125. Smith T, Perry A. A sibling support group for brothers and sisters of children with autism. J Dev Disabil. 2004;11:77–88.
126. Perry A. What it's like to have a brother or sister with a developmental disorder. Toronto: Thistletown Regional Centre; 1989.
127. Spector V, Charlop MH. A sibling-mediated intervention for children with autism spectrum disorder: Using the Natural Language Paradigm (NLP). J Autism Dev Disord. 2018;48:1508–22.
128. Stewart DA, Benson GT, Lindsey JD. A unit plan for siblings of handicapped children. Teach Except Child. 1987;19:24–8.
129. Stewart KK, Carr JE, LeBlanc LA. Evaluation of family-implemented behavioral skills training for teaching social skills to a child with Asperger's disorder. Clin Case Stud. 2007;6:252–62.
130. Kelley ML, Heffer RW, Gresham FM, Elliott SN. Development of a Modified Treatment Evaluation Inventory. J. Psychopathol. Behav. Assess. 1989.
131. Swenson-Pierce A, Kohl FL, Egel AL. Siblings as home trainers: A strategy for teaching domestic skills to children. J Assoc Pers with Sev Handicap. 1987;12:53–60.
132. Trent JA, Kaiser AP, Wolery M. The use of responsive interaction strategies by siblings. Topics Early Child Spec Educ. Thousand Oaks, California: Sage Publications Inc.; 2005;25:107–18.
133. Trent-Stainbrook A, Kaiser AP, Frey JR. Older siblings' use of responsive interaction strategies and effects on their younger siblings with down syndrome. J Early Interv. 2007;29:273–86.
134. Tsao LL, Odom SL. Sibling-mediated social interaction intervention for young children with autism. Topics Early Child Spec Educ. 2006;26:106–23.
135. Tsao LL, McCabe H. Why won't he play with me?: Facilitating sibling interactions. Young Except Child. 2010;13:24–35.
136. Tsao LL. Brothers as playmates for their siblings with developmental disabilities: A multiple-baseline design study. Child Youth Care Forum. 2020;49:409–30.
137. Walton KM, Ingersoll BR. Evaluation of a sibling-mediated imitation intervention for young children with autism. J Posit Behav Interv. 2012;14:241–53.
138. Weinrott MR. A training program in behavior modification for siblings of the retarded. Am J Orthopsychiatry. 1974;44:362–75.
139. Williams PD, Hanson S, Karlin R, Ridder L, Liebergen A, Olson J, et al. Outcomes of a nursing intervention for siblings of chronically ill children: a pilot study. J Soc Pediatr Nurses. 1997;2:127–37.
140. Sahler OJZ, Carpenter PJ. Evaluation of a camp program for siblings of children with cancer. Arch Pediatr Adolesc Med. 1989;143:690.
141. Williams PD, Williams AR, Graff JC, Hanson S, Stanton A, Hafeman C, et al. A community-based intervention for siblings and parents of children with chronic illness or disability: The ISEE study. J Pediatr. 2003;143:386–93.
142. Harter S. Manual for the social support scale for children. Denver, CO: University of Denver; 1985.
143. Harter S. Manual for the self-perception profile for children. Denver, CO: University of Denver; 1985.
144. Williams PD, Williams AR, Hanson S, Gruff C, Ridder L, Curry H, et al. Maternal mood,

family functioning, and perceptions of social support, self-esteem, and mood among siblings of chronically ill children. Child Heal Care. 1999;28:297–310.
145. Eyberg SM, Robinson EA. Conduct problem behavior: Standardization of a behavioral rating. J Clin Child Psychol. 1983;12:347–54.
146. Robinson EA, Eyberg SM, Ross AW. The standardization of an inventory of child conduct problem behaviors. J Clin Child Psychol. 980;9:22–8.
147. Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, et al. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Front Rehabil Sci. Frontiers; 2021;2:52.
148. Lummer-Aikey S, Goldstein S. Sibling adjustment to childhood chronic illness: An integrative review. J Fam Nurs. 2021;27:136–53.
149. Lynam A, Smith MM. Sibling involvement in interventions for children with a disability: a systematic review. Disabil Rehabil. 2021;0:1–11.
150. Lu Y, Douglas SN, Bagawan A, Hauck JL. Using neurotypical siblings as intervention agents to guide individuals with Autism Spectrum Disorders: A systematic review. Res Autism Spectr Disord. 2021;89:1–16.
151. Knox M, Bigby C. Moving towards midlife care as negotiated family business: Accounts of people with intellectual disabilities and their families “Just getting along with their lives together.” Int J Disabil Dev Educ. 2007;54:287–304.
152. Davys D, Mitchell D, Haigh C. Futures planning, parental expectations and sibling concern for people who have a learning disability. J Intellect Disabil. 2010;14:167–83.
153. Leane M. “I don’t care anymore if she wants to cry through the whole conversation, because it needs to be addressed”: Adult siblings’ experiences of the dynamics of future care planning for brothers and sisters with a developmental disability. J Appl Res Intellect Disabil. 2020;33:950–61.
154. Davys D, Mitchell D, Haigh C. Futures planning - adult sibling perspectives. Br J Learn Disabil. 2015;43:219–26.
155. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Can J Community Ment Heal. Canadian Periodical for Community Studies; 2014;33:45–61.
156. Rosenbaum PL, Novak-Pavlic M. Parenting a child with a neurodevelopmental disorder. Curr Dev Disord Reports. 2021;1–7.
157. Hodapp RM, Sanderson KA, Meskis SA, Casale EG. Adult siblings of persons with intellectual disabilities: Past, present, and future. Int Rev Res Dev Disabil. 2017;53:163–202.
158. Peters MDJ, Marnie C, Tricco AC, Pollock D, Munn Z, Alexander L, et al. Updated methodological guidance for the conduct of scoping reviews. JBI Evid Implement. 2021;19:3–10.
159. Munn Z, Peters MDJ, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. BMC Med Res Methodol. 2018;18:143.
160. Smits D-W, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. Res Involv Engagem. 2020;6:30.
161. Mulvale G, Robert G. Special issue: Engaging vulnerable populations in the co-production

of public services. Int J Public Adm. 2021;44:711–4.
162. Meltzer A. What is ‘sibling support’? Defining the social support sector serving siblings of people with disability. 2021;291.
163. Meltzer A. The operation of the social support sector serving siblings of people with disabilities: A cross-country analysis. Soc Policy Adm. 2021;1–15.
164. Nuri RP, Batorowicz B, Aldersey HM. Family support and family and child-related outcomes among families impacted by disability in low- and middle-income countries: A scoping review. J Child Health Care. 2020;24:637–54.

Supplementary File 1: Updated data extraction instrument

Study Information			
Date form completed	Identify the date that the form was completed (dd/mm/yyyy).		
Name of person extracting data	Identify the reviewer who extracted the data for this form.		
Study title	Identify the full title and subtitle.		
Authors	Identify all authors as follows: "last name, initial of first name."		
Author affiliations	List affiliations of all authors (e.g., program, department, institution).		
Country	Identify the country where the study was conducted.		
Study funding source	Describe the funding source for the study and role of funders.		
Possible conflicts of interest	Identify possible conflicts of interest declared by authors.		
Population and Setting			
Participant inclusion criteria	Describe the participant inclusion criteria.		
Participant exclusion criteria	Describe the participant exclusion criteria.		
Participant characteristics	**Total number**	**Number of males**	**Number of females**
	Age (years)		
	Education level		
	Race/ethnicity		
	Diagnosis of participant's siblings		
	Birth order between siblings with and without a neurodisability		
	Description of other participants who are not siblings (if applicable)		
Methods of participant recruitment	Identify the different methods of participant recruitment (e.g., social media, word of mouth, participant referral).		
Setting context	Describe the environment that the program was implemented (e.g., community, college, university, after school).		
Methods			
Study aim	Identify the study aims.		
Type of methodology	Describe the methodology that was used.		
Study steps	Describe the steps to conduct the study.		

Data collection methods	Identify the methods used for data collection (e.g., self-reports, assessment tools, surveys, interviews).
Analysis methods	Identify the methods of analysis (e.g., statistical analyses, qualitative analytic techniques).
Program Characteristics	
Name	Identify the full name and/or abbreviation.
Objective(s) for siblings with a neurodisability	State the objectives of the program for siblings with a neurodisability.
Objective(s) for siblings without a neurodisability	State the objectives of the program for siblings without a neurodisability (e.g., training component).
Duration and frequency	Identify the number and length of sessions, as well as the frequency of sessions.
Mode of delivery	Identify the mode of delivery (e.g., online, in-person).
Activities	Identify the activities that were provided in the programs (e.g., weekly meetings, educational workshops, training activities, follow-up activities).
Sustainability	Describe how the program will be sustained (e.g., activities after the study ends).
Developers	Describe the people who developed the program (e.g., number of developers, background, qualifications, skill level).
Facilitators	Describe the people who facilitated the program (e.g., number of facilitators, background, qualifications, education level).
Participants' needs	Describe the needs (e.g., accessibility and accommodations) of participants in their roles as a sibling while participating in the program.
Participants' goals	Describe the self-reported goals of participants in the program and the progress towards achieving these goals.
Study Results	
Outcomes	Report on the outcomes that were measured in the study.
Key findings for the siblings with a neurodisability	Describe the key findings for the siblings with a neurodisability (e.g., a summary in the discussion section).
Key findings for the siblings without a neurodisability	Describe the key findings for the siblings without a neurodisability (e.g., a summary in the discussion section).
Study reference	Provide the full reference for the study.
Other studies of interest	Identify other studies of interest based on the reference list.

Supplementary File 2. Description of participant characteristics.

Study	**Participant inclusion criteria**	**Total number**	**Number of males**	**Number of females**	**Mean age (SD; standard deviation, range) in years**	**Diagnosis of participant's siblings**	**Birth order between typically developing (TD) compared to the sibling with a neurodisability**	**Description of other participants who are not siblings (if applicable)**
Akers et al. 2018 (1)	Siblings of child who: a) previously or currently attending a university-based behavioral preschool (also known as diagnosis of autism spectrum disorder); b) ability to emit at least 3 word phrases; c) had a generalized imitation repertoire; d) engaged in low levels of destructive behavior; and e) played with toys appropriately but rarely commented during play.	3	1	2	10 (3.27, 6-14)	Autism spectrum disorder	All TD siblings were older than the sibling with autism spectrum disorder	Not applicable
Brouzos et al. 2017 (2)	Siblings of a child with autism spectrum disorder.	38	Experimental group: 10 boys Control group: 8 boys	Experimental group: 12 girls Control group: 8 girls	6-15	Autism spectrum disorder.	Not listed	Not applicable
Burke et al. 2020 (3)	Not mentioned. Siblings of individuals with intellectual and developmental disabilities.	21	6	15	36 (21-67)	Intellectual and developmental disability, which includes intellectual disability, autism	Not listed	Not applicable

						spectrum disorder, cerebral palsy, Down syndrome		
Celiberti et al. 1993 (4)	Siblings who are typically developing at least five years old and not more than one year younger or six years older than the child with autism spectrum disorder. The child with autism spectrum disorder had to engage in little, if any, cooperative play. Siblings should have no known behavioral or academic problems.	3	0	3	8.72 (1.09, 8.16-10.25)	Autism spectrum disorder	TD siblings were older	Not applicable
Chu et al. 2012 (5)	Participants who were typically developing were recruited from similar neighbourhoods to the participants with autism spectrum disorder. They were eligible to participate in the study if they had no history or ongoing medical treatment and met the inclusion criteria for each group. For the sibling-assisted group, both the child with autism spectrum disorder and their typically developing sibling were recruited if they (a) were healthy, (b) were between ages 7 and 12 years old, (c) lived together, and (d)	7	2	5	7.33 (2.41)	Autism spectrum disorder	Not listed	Participants in the peer-assisted and control group

	were able to follow instructions and the requirements of the sibling-assisted aquatic program. For the peer-assisted group and control group of typically developing children, they were recruited if they were (a) healthy, (b) within 7-12 months of age as the other participants had to be, and (c) able to follow the requirements of either the peer-assisted aquatic program or control instructions.							
Clark et al. 1989 (6)	Not mentioned. Siblings of a child with autism spectrum disorder, which was assessed as meeting Diagnostic and Statistical Manual of Mental Disorders, Third Edition (7), criteria for autism spectrum disorder by two independent diagnosticians.	3	1	2		Autism spectrum disorder	Pair 1: TD sibling younger Pair 2: TD sibling older Pair 3: TD sibling older	Parents completed questionnaires
Coe et al. 1991 (8)	Not listed.	2	2	0	10 (1, 9-11)	Autism spectrum disorder	TD siblings were older	Not applicable
Colletti et al. 1977 (9)	Not listed.	3	2	1	11 (0.82, 10-12)	Severe neurological impairment and autism spectrum disorder	TD siblings were older	Parents were trained to be observers of their children's performance
Craft et al. 1990 (10)	Siblings of children with cerebral palsy, ages 4-17 years old, were available	31	15	16	9.58 (3.72, 4-17)	Cerebral palsy	A mixture of older and older and	Not applicable

	for pre-testing, intervention and post-testing, and had given their consent.						younger TD siblings	
Crouthamel 1988 (11)	Not listed.	12	9	3	7-13	Developmental disability	Not listed	Networking community professionals were involved in a separate part of the project
D'Arcy et al. 2005 (12)	Siblings between 8 and 10 years with a sibling with a disability.	16	11	5	8-10	Physical or intellectual disability, or a combination of both	50% older TD siblings, 50% younger TD siblings	Not applicable
Daffner et al. 2020 (13)	Not listed. However, the participant inclusion criteria was provided for the child with attention deficit hyperactivity disorder. The typically developing sibling had to be at least 7 years old, with an age gap between the siblings that could not exceed 6 years.	3	1	2	10.25 (1.43, 8.33-11.75)	Attention deficit hyperactivity disorder	TD siblings were older	Parent completed questionnaires.
Doleys et al. 1975 (14)	Not listed.	1	0	1	19	Intellectual and developmental disorder	TD siblings were older	Not applicable.
Douglas et al. 2018 (15)	Siblings with typical development, between 7 to 15 years old, has a sibling with a communication disability who requires the use of alternative communication such as sign, gestures, computer systems, or picture	3	1	2	10.44 (3.16, 8.08-14.92)	Speech and motor delay and an emotional disability; Down syndrome; Noonan Syndrome	TD siblings were older	Not applicable.

	symbols. When there was more than one typically developing sibling who met the eligibility requirements, the parent was instructed to select the typically developing sibling who played most frequently with the child with complex communication needs.					characterized by developmental delays, hypotonia, and vision problems		
Dyson 1998 (16)	Siblings of a sibling with a disability.	40	24	16	7.5-12	Intellectual and developmental disability, autism spectrum disorder, attention deficit disorders sensory impairment, physical disability, learning disabilities and communication disorders, developmental delay, and unspecified disability	Not listed.	Not applicable.
Evans et al. 2001 (17)	Not listed.	28	9	19	6-12	Learning disabilities	Not listed.	Not applicable.
Ferraioli et al. 2011 (18)	Siblings of a sibling with a diagnosis of autism spectrum disorder according to Diagnostic	4	4	0	7.33 (0.96, 6.0 – 8.33)	Autism spectrum disorder	TD siblings were older	Not applicable

	and Statistical Manual of Mental Disorders, Fourth Edition (19), criteria, as determined by an unaffiliated professional.							
Fjermestad et al. 2020 (20)	Sibling of an individual with a neurodevelopmental disorder.	52	66%	44%	12.70 (2.70, 8-21)	Learning problems, intellectual disability, autism spectrum disorder, attention deficit hyperactivity disorder, developmental delay, or Down Syndrome	Not listed	Parents as participants in the program
Fjermestad et al. 2020 (21)	1) Being the sibling of a child diagnosed with a neurodevelopmental disorder who is aged 0 to 18 years and who receives specialist and/or municipal health services; 2) ages 8-16 years; and 3) one parent able to attend the intervention.	Anticipated enrollment of 288 participants.	Not applicable	Not applicable	Not applicable	Neurodevelopmental disorder	Not applicable.	Parents as participants in the program
Gettings et al. 2015 (22)	Not listed. Siblings of patients being treated at the Centre for Interventional Pediatric Psychopharmacology, a national specialist Child and Adolescent Mental Health Service in the UK. All patients had complex	6	1	5	8-13	Autism spectrum disorder, ADHD, mood disorder, obsessive compulsive disorder, Down's syndrome,	Siblings without a neurodisability were younger than siblings with a neurodisability.	All siblings were accompanied by parents. Five mothers and one father took part.

	neurodevelopmental disorders involving at least two co-morbid conditions such as autism spectrum disorder, attention deficit hyperactivity disorder, obsessive compulsive disorder, oppositional defiant disorder or anxiety disorders.					oppositional defiant disorder, visual impairment, multiple anxiety disorders or phobias		
Granat et al. 2012 (23)	Siblings in families of children registered at an outpatient habilitation centre in mid-Sweden, ages 8-12 years old with no diagnosed disability.	54	21	33	8-12 y	Attention deficit hyperactivity disorder, Asperger syndome, autism spectrum disorder, physical disability or intellectual disability.	31 TD siblings were older	Not applicable.
Hancock et al. 1996 (24)	The sibling was (a) at least 8 years of age, (b) chronologically older than the sibling with a neurodisability, and (c) willing to participate in the study. The sibling with a neurodisability was (a) between the ages of 4 and 8 years, (b) functioning in the mild to moderate range of an intellectual and developmental disorder, (c) spontaneously using one- to three-word utterances, and (d) socially	3	0	3	10.67 (1.89, 8-12)	Cerebral palsy, developmental delay of unknown origin, William's syndrome	TD siblings were older	Not applicable

	responsive to initiations made by his sibling.							
Hayden et al. 2019 (25)	Not listed. Sibling of an individual with a disability.	55	Number not listed, 45.5%	Number not listed. 54.5%	9.18 years (7-11)	Autism, Down syndrome, hearing impairments or chronic medical condition	Not listed	Not applicable
James et al. 1986 (26)	Not listed. Sibling of an individual with a disability.	3	1	2	6.83-8.08	Cerebral palsy and intellectual disability	TD siblings were older .	TD peers nonhandicapped peers friends of the siblings participated, and parent ratings were collected
Jones et al. 2020 (27)	Sibling of an individual with autism spectrum disorder.	54 (24 in the control group, 30 in the support group)	24	30	8.31 years (3.52)	Autism spectrum disorder	Control group: 15 TD siblings were older, 13 were younger, and 2 were of the same age. Support group: 12 TD siblings were older than their siblings with autism spectrum disorder, 9 were younger, and 3 were of the same age.	Parents completed measures
Kryzak et al. 2015 (28)	Typically developing sibling (i.e., with no known diagnosis of autism spectrum disorder) of a child who has a diagnosis	15	9	6	4-14 years old	Autism spectrum disorder, which included	Not listed.	Sibling with autism spectrum disorder.

	of autism spectrum disorder from an outside source (per DSM-IV-TR; American Psychiatric Association, 2000).					pervasive developmental disorder not otherwise specified, autism, Asperger's, or autism spectrum diagnoses		
Kryzak et al. 2017 (29)	Average performance on the Stanford-Binet Intelligence Scales - Fifth Edition; and nonclinical scores on the parent-completed Child Behavior Checklist. Sibling of a sibling with autism spectrum disorder, parent report of a diagnosis of autism spectrum disorder made by practitioners not associated with the study.	4	1	3	8.5 (2.60, 6-12)	Autism spectrum disorder	Not listed	Four typically developing sibling dyads participated to provide a comparison with interactions between TD sibling and sibling with autism spectrum disorder dyads for social validity purposes.
Lewandowski et al. 2014 (30)	Not listed. TD sibling of a child with autism spectrum disorder.	1	1	0	6.17	Autism spectrum disorder or Asperger syndrome	TD sibling was younger	The children's birth mother completed measures
Lobato et al. 1985 (31)	Not listed. Sibling of a sibling with a neurodisability.	1	0	1	21	Down's Syndrome	TD sibling was older	Discussions were held with the child's parents, two sisters, and maternal grandmother.

Lobato 1985 (32)	Not listed. TD sibling of a sibling with a disability.	6	3	3	5.33 (3.75-7)	Hearing loss, left hemiplegia due to a stroke, cerebral palsy, Down syndrome, and intellectual disorder	5 TD siblings were older	Parent recordings of sibling interactions at home
Lobato et al. 2002 (33)	Not listed. TD siblings, ages 8 to 13 years old, of a sibling with a chronic illness/developmental disability.	54	24	30	9.8 (8-13)	Physical disabilities, autism spectrum disorders, intellectual, medical disorders or combined psychiatric and learning disorders	57% of TD siblings were older	Parents participated in the program and completed measures
Lobato et al. 2005 (34)	Not listed. TD siblings (ages 4-7 years) of a sibling with a chronic illness or developmental disability	43	17	26	5.7 (4-7)	Autism spectrum disorders including Asperger's disorder, intellectual disability, physical disabilities such as cerebral palsy, medical disorders such as cancer, or dual psychiatric and learning	51% of the TD siblings were older	Parents participated in the program and completed measures

						disorders such as Tourette's		
McCullough et al. 2011 (35)	Not listed.	3	1	2	Not listed.	Autism spectrum disorder and intellectual disability with limited verbal abilities.	Not listed	Not applicable
McLinden et al. 1991 (36)	Sibling of a sibling with a disability.	6	1	5	9.17	Intellectual disability, physical disability, or multiple disabilities	Not listed.	Parents completed a questionnaire, and mothers were interviewed
Miller et al. 1976 (37)	Not listed. Sibling of a sibling with a neurodisability.	8	5	3	17.25 (1.92, 15-20) *Note: ages of siblings in the first case study were not provided).	Congenital facial anomaly and slow early development, autism spectrum disorder, intellectual disability	TD siblings were older	Parents were participants in the program
Neff et al. 2017 (38)	Not listed. Sibling of a sibling with autism spectrum disorder.	3	1	2	4.67 (0.94, 4-6)	Autism spectrum disorder	Twins, one TD sibling was older, and one TD sibling was younger	Not applicable
Oppenheim-Leaf et al. 2012 (39)	Not listed. TD sibling of a sibling with autism spectrum disorder.	3	2	1	4.67 (0.47, 4-5)	Autism spectrum disorder	Two TD siblings were older and one TD sibling was younger	Not applicable
Özen 2015 (40)	Pre-requisite skills that TD siblings should have are: (i) volunteer for participation in the study,	3	Not listed.	Not listed.	9.67 (0.94, 9-11)	Autism spectrum disorder	All TD siblings were older	Mothers were asked to complete a questionnaire

	(ii) be a student in primary school, (iii) be independent in using an iPad, (iv) be able to play the selected iPad game after one or two rehearsals, (v) be able to understand the visual material and verbal presentation regarding the sibling training session, (vi) be able to interact socially with peers and adults, and (vii) be available to participate in the study at least twice a week. Playing with the iPad should be one of the favorite leisure activities of the siblings as well. The following pre-requisite skills were for the siblings with autism spectrum disorder: (i) have a diagnosis of autism spectrum disorder, (ii) be able to follow directions, (iii) being able to pay attention to visual or verbal stimulus for at least 5 minutes, (iv) be able to keep eye contact, (v) be able to watch a scene on the computer or iPad for at least 2 minutes, (vi) not have any experience of systematical training with the iPad, and (vii) be able to slide their index finger							

	on the iPad screen and press with their finger.							
Phillips 1999 (41)	Siblings of children with identified developmental disabilities.	180	72	108	11.3 (9-12)	Intellectual disability	Not listed	Not applicable
Roberts et al. 2015 (42)	Sibling had to be aged between 8 and 12 years with 6 months allowance on either side; both sibling and parent consent had to be obtained; and siblings themselves could not have a significant disability or chronic health condition.	42 (22 in intervention group, 20 in waitlist control group)	25	17	9.3 (1.38, 7.5-12.5)	autism spectrum disorder, Angelman's syndrome, Down syndrome, Phelan-McDermid syndrome, global developmental delay, pervasive developmental disorder, intellectual disability, and optic nerve hypoplasia	21 TD siblings were older	Parents completed outcome measures on behalf of participants. The parent component of SibworkS consisted of Parent Information Sheets provided in the manual. These summarized the the main theme of each session and the rationale for exploring it. They also offer ‘Parent Tips’, which are a mixture of practical tips and advice for encouraging further discussion about the session theme.

Roberts et al. 2016 (43)	Siblings aged between 8 and 12 with 6 months allowance either side, could not have a disability, and have a sibling with a disability (i.e., special requirements for their care as a result of a physical or developmental challenge).	36	14	22	9.14 (1.25, 7-13)	Autism spectrum disorder, Crohn's disease, epileptic encephalopathy, attention-deficit hyperactivity disorder, global developmental delay, low muscle tone, dyslexia, dyspraxia, cleft lip, anxiety, depression, oppositional defiance disorder, sensory processing disorder, dysgraphia, Angelman's syndrome, Down’s syndrome, Phelan McDermid syndrome, global developmental delay, pervasive developmental disorder,	Not listed.	Parent component, in which parent information sheets were provided in the manual. These summarize the main theme of each session (1–6 above) and the rationale for its incorporation into the program. They also offer ‘Parent Tips’, which are a combination of practical tips as well as advice for encouraging further discussion about the session themes.

						intellectual disability and optic nerve hypoplasia		
Rye et al. 2018 (44)	Siblings of an individual with a disability known to the Ealing Service for Children with Additional Needs.	4	0	4	8-13	Disabilities, including autism spectrum disorder, severe learning disability, chromosomal deletion, cerebral palsy and epilepsy	Not listed.	Not applicable.
Schreibman et al. 1983 (45)	Siblings of a sibling with autism spectrum disorder that was diagnosed by at least two outside agencies not associated with the research projects.	3	1	2	13, 11, 8	Autism spectrum disorder	TD siblings were older	Not applicable
Sheikh et al. 2019 (46)	Siblings had to be typically developing, have a sibling with autism spectrum disorder diagnosed by a professional not associated with this research, with a parent interested in parent-sibling training.	3	1	2	5 (0.82, 4-6)	Autism spectrum disorder	One sibling-dyad were twins, and two sibling-dyads the TD siblings were younger	Parents were involved in the parent-sibling training that involved the whole family
Smith et al. 2004 (47)	Not listed. Siblings of families who attended the TRE-ADD (Treatment, Research, and Education for Autism and Developmental Disorders) program at Thistletown Regional Centre, of a	26	12	~~14~~	10.63 (2.13, 6.58-16.25)	Autism spectrum disorder or related disorder (e.g., pervasive developmental disorder, Rett	14 TD siblings were older, 12 TD siblings were younger	Parents completed questionnaires

	sibling with autism or related disorder (e.g., pervasive developmental disorder, Rett disorder, or developmental delay).					disorder, or developmental delay)		
Spector et al. 2018 (48)	Siblings had to be typically developing and have a sibling with autism spectrum disorder diagnosed by two licensed psychologists from two independent agencies not affiliated with this study according to the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (49) and also displayed a speech deficit.	3	1	2	9 (1.63, 7-11)	Autism spectrum disorder and speech deficit	TD siblings were older	Not applicable
Stewart et al. 1987 (50)	Not a study with participants. However, the program is for siblings of individuals with a disability.	Not applicable.	Not applicable.	Not applicable.	Not applicable.	Disability.	Not applicable.	Not applicable.
Stewart et al. 2007 (51)	Not listed. Sibling of a sibling with autism spectrum disorder (specifically Asperger's disorder)	1	0	1	10	Asperger's disorder and attention-deficit/hyperactivity disorder	TD sibling was the same age (both part of triplets)	The sibling participant assisted mother during training.
Swenson-Pierce et al. 1987 (52)	Older TD siblings of a sibling with a disability and a need for the sibling with a disability to acquire domestic skills.	3	1	2	12 (1.41, 10-13)	Intellectual disability, seizures, Down's syndrome, microcephaly, upper extreme spasticity	TD siblings were older	Not applicable
Trent et al. 2005 (53)	Siblings between 6-12 years old, were	2	0	2	8 (1, 7-9)	Down syndrome	TD siblings were older	Not applicable

	chronologically older than the siblings with Down syndrome, and the siblings with Down syndrome were between 5-11 years old with significant language delays.							
Trent-Stainbrook et al. 2007 (54)	Siblings between 6-12 years old, have a younger sibling with Down syndrome between 5-11 years old.	3	1	2	9.33 (0.47, 9-10)	Down syndrome	TD siblings were older	Not applicable
Tsao et al. 2006 (55)	Not listed. Siblings were typically developing, between 4 to 11 years of age of a sibling between 3 and 6 years of age diagnosed with autism or another autistic spectrum disorder (i.e., pervasive developmental disorder, not otherwise specified) by a physician, psychologist, or diagnostic clinic staff member using the criteria from the Diagnostic and Statistical Manual of Mental Disorders-Fourth Edition (19)	4	2	2	6.27 (2.83, 4.50-11.17)	Autism and Asperger syndrome	Two TD siblings were younger, and two TD siblings were older	Parents would stay with their children throughout the experiment. During the baseline, parents were asked to act as they usually did with their children. In the intervention phases, the parents watched how the researcher taught a lesson to the TD sibling and could offer suggestions that might help the TD sibling understand the lesson. Parents

								could also offer verbal prompts to both of siblings in the dyad during the intervention and maintenance phases. At the end of each play session in intervention, parents were asked to offer feedback on the lesson plan.
Tsao et al. 2010 (56)	N/A	N/A	N/A	N/A	N/A	Disabilities	N/A	N/A
Tsao 2020 (57)	Not listed. TD brothers of a brother with a developmental disability evaluated by a certified psychologist or clinician.	3	3	0	5.11 (1.54, 3.33-7.08)	Developmental disabilities, including autism spectrum disorder	Two TD siblings were older and one TD sibling was younger	Parents were present at each session but did not engage in the interactions
Walton et al. 2012 (58)	Not listed. TD sibling of a sibling with autism spectrum disorder diagnosed by a professional with expertise in autism and exceeded the cut-off scores for "autism spectrum" on the social, communication, and social + communication algorithms of the Autism Diagnostic Observation Schedule-Module 1 (59).	6	2	4	9.5 (1.71, 8-13)	Autism spectrum disorder	TD siblings were older	Parents reported on the satisfaction with the program

Weinrott 1974 (60)	Siblings ages 10-18 of a sibling with an intellectual disability who were living in the Boston area, since pre-camp training would be held at Harvard University and The Walter E. Fernald State School.	18	Not listed.	Not listed.	between 10 and 18 years old.	Intellectual disability	Not listed.	Parents participated in the training program.
Williams et al. 1997 (61)	Not listed. Siblings of children with diagnoses of cancer, cystic fibrosis, diabetes, or spina bifida.	22	9	13	8.5 years (5.57)	Cancer, cystic fibrosis, diabetes, or spina bifida	Not listed.	Parents attended a discussion about issues related to sibling adjustment.
Williams et al. 2003 (62)	TD sibling who was closest in age to the sibling with a neurodisability.	79	41	38	11.1 years (2.2)	Cystic fibrosis, diabetes, spina bifida, cancer, and developmental disabilities (which included Down syndrome, autism, traumatic brain injury, or cerebral palsy)	Not listed.	Parents participated in sessions and completed questionnaires.

*Note 1: For the purpose of this review, the gender of participants was reported instead of sex. The reporting of gender aligns with its definition as a socially constructed role (63), and siblings may choose to have different roles while participating in programs.

**Note 2: The mean age and standard deviation was manually calculated, if the authors of included studies provided all available data for each individual participant.

References for Supplementary File 2

1. Akers JS, Higbee TS, Pollard JS, Reinert KS. Sibling-implemented script fading to promote play-based statements of children with autism. Behav Anal Pract. 2018;11(4):395–9.
2. Brouzos A, Vassilopoulos SP, Tassi C. A psychoeducational group intervention for siblings of children with autism spectrum disorder. J Spec Gr Work. 2017;42(4):274–98.
3. Burke MM, Lee CE, Carlson SR, Arnold CK. Exploring the preliminary outcomes of a sibling leadership program for adult siblings of individuals with intellectual and developmental disabilities. Int J Dev Disabil. 2018;0(0):1–8.
4. Celiberti DA, Harris SL. Behavioral intervention for siblings of children with autism: A focus on skills to enhance play. Behav Ther. 1993;24(4):573–99.
5. Chu CH, Pan CY. The effect of peer- and sibling-assisted aquatic program on interaction behaviors and aquatic skills of children with autism spectrum disorders and their peers/siblings. Res Autism Spectr Disord. 2012;6(3):1211–23.
6. Clark ML, Cunningham LJ, Cunningham CE. Improving the social behavior of siblings of autistic children using a group problem solving approach. Child Fam Behav Ther. 1989;11(1):19–33.
7. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 3rd ed. 1980.
8. Coe DA, Matson JL, Craigie CJ, Gossen MA. Play skills of autistic children: Assessment and instruction. Child Fam Behav Ther. 1991;13(3):13–40.
9. Colletti G, Harris SL. Behavior modification in the home: Siblings as behavior modifiers, parents as observers. J Abnorm Child Psychol. 1977;5(1):21–30.
10. Craft MJ, Lakin JA, Oppliger RA, Clancy GM, Vanderlinden DW. Siblings as change agents for promoting the functional status of children with cerebral palsy. Dev Med Child Neurol. 1990;32(12):1049–57.
11. Crouthamel CS. Siblings of handicapped children: A group support program. Early Child Dev Care. 1988;37:119–31.
12. D'Arcy F, Flynn J, McCarthy Y, O'Connor C, Tierney E. Sibshops. An evaluation of an interagency model. J Intellect Disabil. 2005;9(1):43–57.
13. Daffner MS, DuPaul GJ, Kern L, Cole CL, Cleminshaw CL. Enhancing social skills of young children with ADHD: Effects of a sibling-mediated intervention. Behav Modif. 2020;44(5):698–726.
14. Doleys DM, Slapion MJ. The reduction of verbal repetitions by response cost controlled by a sibling. J Behav Ther Exp Psychiatry. 1975;6(1):61–3.
15. Douglas SN, Kammes R, Nordquist E, D'Agostino S. A pilot study to teach siblings to support children with complex communication needs. Commun Disord Q. 2018 Feb;39(2):346–55.
16. Dyson LL. A support program for siblings of children with disabilities: What siblings learn and what they like. Psychol Sch. 1998;35(1):57–65.
17. Evans J, Jones J, Mansell I. Supporting siblings: Evaluation of support groups for brothers and sisters of children with learning disabilities and challenging behaviour. J Learn Disabil. 2001;5(1):69–78.
18. Ferraioli SJ, Harris SL. Teaching joint attention to children with autism through a sibling-mediated behavioral intervention. Behav Interv. 2011;26(4):261–81.
19. American Psychiatric Association. Diagnostic and statistical manual of mental disorders.

4th ed. Washington, DC: American Psychiatric Association; 2000.

20. Fjermestad K, Pat P, Dearozet S, Vatne T, Hafting M, Jegannathan B. Manual-Based Group Intervention for Siblings and Parents of Children with Neurodevelopmental Disorders in Cambodia. J Dev Phys Disabil. 2021;33(5):839–56.
21. Fjermestad KW, Silverman WK, Vatne TM. Group intervention for siblings and parents of children with chronic disorders (SIBS-RCT): study protocol for a randomized controlled trial. Trials. 2020;21(1):1–12.
22. Gettings S, Franco F, Santosh PJ. Facilitating support groups for siblings of children with neurodevelopmental disorders using audio-conferencing: A longitudinal feasibility study. Child Adolesc Psychiatry Ment Health. 2015;9(1):1-15.
23. Granat T, Nordgren I, Rein G, Sonnander K. Group intervention for siblings of children with disabilities: a pilot study in a clinical setting. Disabil Rehabil. 2012;34(1):69–75.
24. Hancock TB, Kaiser AP. Siblings' use of milieu teaching at home. Topics Early Child Spec Educ. 1996;16(2):168–90.
25. Hayden NK, McCaffrey M, Fraser-Lim C, Hastings RP. Supporting siblings of children with a special educational need or disability: An evaluation of Sibs Talk, a one-to-one intervention delivered by staff in mainstream schools. Support Learn. 2019;34(4):404–20.
26. James SD, Egel AL. A direct prompting strategy for increasing reciprocal interactions between handicapped and nonhandicapped siblings. J Appl Behav Anal. 1986;19(2):173–86.
27. Jones EA, Fiani T, Stewart JL, Neil N, McHugh S, Fienup DM. Randomized controlled trial of a sibling support group: Mental health outcomes for siblings of children with autism. Autism. 2020;24(6):1468–81.
28. Kryzak LA, Cengher M, Feeley KM, Fienup DM, Jones EA. A community support program for children with autism and their typically developing siblings: Initial investigation. J Intellect Disabil. 2015;19(2):159–77.
29. Kryzak LA, Jones EA. Sibling self-management: Programming for generalization to improve interactions between typically developing siblings and children with autism spectrum disorders. Dev Neurorehabil. 2017;20(8):525–37.
30. Lewandowski JF, Hutchins TL, Prelock PA, Murray-Close D. Examining the benefit of including a sibling in story-based intervention with a child with Asperger Syndrome. Contemp Issues Commun Sci Disord. 2014;41(Fall):179–95.
31. Lobato D, Tlaker A. Sibling intervention with a retarded child. Educ Treat Child. 1985;8(3):221–8.
32. Lobato D. Brief report: Preschool siblings of handicapped children - Impact of peer support and training. J Autism Dev Disord. 1985;15(3):345–50.
33. Lobato DJ, Kao BT. Integrated sibling-parent group intervention to improve sibling knowledge and adjustment to chronic illness and disability. J Pediatr Psychol. 2002;27(8):711–6.
34. Lobato DJ, Kao BT. Brief report: Family-based group intervention for young siblings of children with chronic illness and developmental disability. J Pediatr Psychol. 2005;30(8):678–82.
35. McCullough K, Simon SR. Feeling heard: a support group for siblings of children with developmental disabilities. Soc Work Groups. 2011;34(3–4):320–9.
36. McLinden SE, Miller LM, Deprey JM. Effects of a support group for siblings of children with special needs. Psychol Sch. 1991;28:230–7.

37. Miller NB, Cantwell DP. Siblings as therapists: a behavioral approach. Am J Psychiatry. 1976;133(4):447–50.
38. Neff ER, Betz AM, Saini V, Henry E. Using video modeling to teach siblings of children with autism how to prompt and reinforce appropriate play. Behav Interv. 2017;32(3):193–205.
39. Oppenheim-Leaf ML, Leaf JB, Dozier C, Sheldon JB, Sherman JA. Teaching typically developing children to promote social play with their siblings with autism. Res Autism Spectr Disord. 2012;6(2):777–91.
40. Özen A. Effectiveness of siblings-delivered iPad game activities in teaching social interaction skills to children with autism spectrum disorders. Kuram ve Uygulamada Egit Bilim. 2015;15(5):1287–303.
41. Phillips RSC. Intervention with siblings of children with developmental disabilities from economically disadvantaged families. Fam Soc. 1999;80(6):569–77.
42. Roberts RM, Ejova A, Giallo R, Strohm K, Lillie M, Fuss B. A controlled trial of the SibworkS group program for siblings of children with special needs. Res Dev Disabil. 2015;43:21–31.
43. Roberts RM, Ejova A, Giallo R, Strohm K, Lillie ME. Support group programme for siblings of children with special needs: predictors of improved emotional and behavioural functioning. Disabil Rehabil. 2016;38(21):2063–72.
44. Rye K, Hicks S, Falconer C. Evaluating a group for young people who have a sibling with a disability. Learn Disabil Pract. 2018 May 15;1.
45. Schreibman L, O’Neill RE, Koegel RL. Behavioral training for siblings of autistic children. J Appl Behav Anal. 1983;16(2):129–38.
46. Sheikh R, Patino V, Cengher M, Fiani T, Jones EA. Augmenting sibling support with parent-sibling training in families of children with autism. Dev Neurorehabil. 2019;22(8):542–52.
47. Smith T, Perry A. A sibling support group for brothers and sisters of children with autism. J Dev Disabil. 2004;11(1):77–88.
48. Spector V, Charlop MH. A sibling-mediated intervention for children with autism spectrum disorder: Using the Natural Language Paradigm (NLP). J Autism Dev Disord. 2018;48(5):1508–22.
49. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Arlington, VA: American Psychiatric Publishing, Inc.; 2013.
50. Stewart DA, Benson GT, Lindsey JD. A unit plan for siblings of handicapped children. Teach Except Child. 1987;19(3):24–8.
51. Stewart KK, Carr JE, LeBlanc LA. Evaluation of family-implemented behavioral skills training for teaching social skills to a child with Asperger’s disorder. Clin Case Stud. 2007;6(3):252–62.
52. Swenson-Pierce A, Kohl FL, Egel AL. Siblings as home trainers: A strategy for teaching domestic skills to children. J Assoc Pers with Sev Handicap. 1987;12(1):53–60.
53. Trent JA, Kaiser AP, Wolery M. The use of responsive interaction strategies by siblings. Topics Early Child Spec Educ. 2005;25(2):107–18.
54. Trent-Stainbrook A, Kaiser AP, Frey JR. Older siblings’ use of responsive interaction strategies and effects on their younger siblings with down syndrome. J Early Interv. 2007;29(4):273–86.
55. Tsao LL, Odom SL. Sibling-mediated social interaction intervention for young children

with autism. Topics Early Child Spec Educ. 2006;26(2):106–23.

56. Tsao LL, McCabe H. Why won't he play with me?: Facilitating sibling interactions. Young Except Child. 2010;13(4):24–35.
57. Tsao LL. Brothers as playmates for their siblings with developmental disabilities: A multiple-baseline design study. Child Youth Care Forum. 2020;49(3):409–30.
58. Walton KM, Ingersoll BR. Evaluation of a sibling-mediated imitation intervention for young children with autism. J Posit Behav Interv. 2012;14(4):241–53.
59. Lord C, Risi S, Lambrecht L, Cook, Jr. EH, Leventhal BL, DiLavore PC, et al. The Autism Diagnostic Observation Schedule—Generic: A Standard Measure of Social and Communication Deficits Associated with the Spectrum of Autism. J Autism Dev Disord. 2000;30(3):205–23.
60. Weinrott MR. A training program in behavior modification for siblings of the retarded. Am J Orthopsychiatry. 1974;44(3):362–75.
61. Williams PD, Hanson S, Karlin R, Ridder L, Liebergen A, Olson J, et al. Outcomes of a nursing intervention for siblings of chronically ill children: A pilot study. J Soc Pediatr Nurses. 1997;2(3):127–37.
62. Williams PD, Williams AR, Graff JC, Hanson S, Stanton A, Hafeman C, et al. A community-based intervention for siblings and parents of children with chronic illness or disability: The ISEE study. J Pediatr. 2003;143(3):386–93.
63. Canadian Institutes of Health Research. What is gender? What is sex? [Internet]. 2020 [cited 2021 Dec 12]. Available from: https://cihr-irsc.gc.ca/e/48642.html

CHAPTER FIVE

TITLE: Protocol of the BEST SIBS study: A qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition

AUTHORS: Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW.

JOURNAL: Journal of Transition Medicine

CITATION: Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Protocol of the BEST SIBS study: a qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition. Journal of Transition Medicine. 2021;3(1). doi: 10.1515/jtm-2021-0004

Abstract

Background: Children and youth with neurodisabilities may experience different challenges during their transition to adulthood, such as pursuing postsecondary education, finding employment, and navigating a new adult health care system. Families, including siblings, have an important role in the process for when youth with neurodisabilities are transitioning to adulthood. Siblings are in a unique position, where they can have different roles such as a friend, mentor, or caregiver. Siblings can offer various supports to their brother or sister with a neurodisability, but they require knowledge and skills for these different supporting roles. Currently, there are limited programs available for siblings to learn how to support their brother or sister with a neurodisability during transition. A first step to develop these sibling support programs is to understand the experiences of siblings of youth with a neurodisability. The purpose of this report is to describe a protocol of a qualitative case study aimed at examining the experiences of siblings including their roles and responsibilities to their brother or sister with a neurodisability during health care transition.

Methods: An exploratory single case study design will be used. An integrated knowledge translation approach to conducting this study will be used by partnering with the Sibling Youth Advisory Council comprised of siblings who have a brother or sister with a disability throughout all study phases. Participants will include siblings (ages 14–40 years old) with a brother or sister (ages 14–21 years old) with a neurodisability in Ontario, Canada. Semi-structured interviews will be conducted, that will be augmented by photo elicitation and drawings of family tree diagrams. Data will be analyzed using reflexive thematic analysis.

Discussion: Findings from this study will be shared with siblings, families, researchers, and the broader community. It is important to understand the roles and responsibilities that siblings are choosing to have to support their brother or sister with a neurodisability, and how these roles may change over time as their sibling is growing up and transitions to adulthood. Siblings may require knowledge and skills in these roles, and an understanding about siblings' experiences in certain roles can help to inform the development of a resource to support siblings during health care transition.

Keywords: disability, health care transition, qualitative study, sibling

Introduction

Youth have numerous opportunities to explore their future interests and goals, including school, work, family, and leisure [1]. These interests and goals are often explored during adolescence, which is a developmental period that occurs from age 12 to 25, where youth are transitioning to adulthood [2]. However, youth with a neurodisability, such as autism spectrum disorder or cerebral palsy, often face biological, social, and emotional challenges when they are in the phase of becoming an adult [3]. These challenges may include pursuing postsecondary education, finding employment, developing long-term relationships, and navigating adult services including health care [3]. Youth with a neurodisability may also experience challenges during health care transition when they need to learn how to navigate adult health care services [3]. Health care transition for youth is defined as the "purposeful, planned movement of adolescents with chronic medical conditions from child-centred to adult-oriented health care" [4]. This experience can be described as 'falling off a cliff', where youth are often unprepared for this transition when their familiar pediatric services abruptly end [5]. Youth might feel this way because they find these new adult services challenging to navigate [5]. In these situations many youth can turn to family members, who have most likely been involved in navigating their care systems, for support during this transition [3].

In order for successful health care transition to occur, youth and families need to be prepared for this process. Siblings are also a part of the family, but their role in health care transition is often unclear. Siblings share a lifelong bond and often understand the unique needs or concerns of their brother or sister with a neurodisability [6]. While growing up, typically developing (TD) siblings can have different roles such as a mentor, role model, or friend to their brother or sister with a disability [7]. Siblings often take on formal and informal roles for their brother or sister with a disability, which were often assumed by the family or based on needs of the family's circumstances [7]. At a young age, siblings may recognize that they need to support their family in different contexts [8]. For example, they may recognize their role in caring for their sibling when a parent or adult caregiver is unavailable [8]. In many families, formal discussions around the role of siblings and expectations for their relationship is not done until long after the transition process [9]. This leads to missed opportunities for engagement in health care, learning opportunities, and anxiety about the future of the sibling relationship and roles. It is essential to understand the multifaceted roles of siblings as they may be present in the lives of their brother or sister for a longer period of time than any other family member [9]. There should be increased awareness and understanding about the potential unique roles that siblings can have during health care transition.

In 2018, the Siblings Needs Assessment Survey collected data from 360 young adults (≥20 years old) in Canada (87.2% respondents from Ontario) who had a brother or sister with a disability [10]. The most commonly identified needs were options for housing and finances to support their brother or sister [10]. Siblings have described concerns for their brother or sister's future such as finding employment or living independently [11]. Siblings might have worries for new responsibilities, such as guardianship or financial responsibilities, when their parents can no longer be the primary caregivers [12]. These concerns can affect the extent to which siblings gradually become involved throughout the lives of their brother or sister with a neurodisability.

Siblings who are developing typically might want to support their brother or sister with a disability, but they require knowledge and skills on how to do this. Siblings identified that they want to connect with others, and share their experiences of their sibling relationship, so that they can learn from each other [8]. A systematic review of 17 studies identified that intervention programs targeting siblings' behaviour and knowledge can lead to improvements on their health and well-being [13]. Other programs, such as the 'Sibshops' are currently conducted across 10 different countries, including the United States and Canada [14]. These Sibshops provide opportunities for siblings to connect with each other, discuss the joys and concerns of siblings, and learn about coping strategies. However, the Sibshops are targeted for TD siblings who are ages 8–13 years old. While there are some SibTeen sessions tailored for adolescents ages 13–17 years old, they are held monthly with recreational activities [15]. Currently, there are limited resources available for siblings who are 14 years and older to support their brother or sister to prepare for health care transition. The Siblings Needs Assessment Survey identified that siblings have a variety of needs and questions about how they can best support their brother or sister with a neurodisability, and there needs to be further understanding about how to support siblings' needs [10].

A critical first step in developing interventions is to supplement the existing literature with new primary research through interviews with key stakeholders [16]. In this proposed study, the target population is the siblings of a brother or sister with a neurodisability who are preparing for and experiencing the process of health care transition. Siblings also need to be prepared in their roles during health care transition. We need to understand the siblings' experiences, and specifically their roles and responsibilities during adolescence, that will inform the possible gaps and topics to address in the development of future resources or tools to support youth and young adult siblings of individuals with a neurodisability.

Objectives

The purpose of this study protocol is to describe a qualitative case study: BrothErs and

Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study. This study has the two objectives, to: 1) deepen our understanding of sibling roles, including any functions associated with health care transitions; and 2) identify the siblings' responsibilities in their relationship with their brother or sister with a neurodisability.

It is important to have a holistic understanding of siblings' experiences including how certain events shape their relationships with their siblings with a neurodisability. For example, siblings might decide to change their own plans to spend time with their sibling with a neurodisability when their parents were not able to [17]. As siblings become young adults, they might choose to live at home in order to offer support to their sibling with a neurodisability and family [9]. The support that siblings might offer to their brother or sister with a neurodisability can depend on commitments with their family, social, and work [9]. An important event in the relationship between siblings is health care transition, which is a process and this study focuses on the 'preparation' and 'journey' phases [18]. This study will provide an opportunity to share stories and raise awareness about siblings' experiences and the roles and responsibilities in the present and future while the process of transition is taking place. These stories can then be shared with other individuals, such as families and service providers (e.g., health care) through the co-creation of knowledge translation products with sibling partners, such as infographics, conference presentations and workshops, and videos to be posted on social media. These stories will also be posted as a summary on the BEST SIBS Study website after the study is completed.

Integrated knowledge translation

Integrated knowledge translation is an approach to doing research with knowledge users, such as patients and families, as equal partners with researchers throughout the study [19].

Siblings with the lived experiences of having a brother or sister with a disability have often described the importance of being involved in research [20]. In 2018, we established an advisory council of siblings who have a brother or sister with a disability called the Sibling Youth Advisory Council (SibYAC) as a research partner in this doctoral study (LN). Members of the SibYAC were recruited by word-of-mouth who were already connected with the research team, such as through their involvement with other advisory councils or with the research centre. The SibYAC is currently comprised of six young adults (ages 21–27 years old), with five sisters and one brother. There are five members living in Ontario, Canada and one member living in Alberta, Canada. All SibYAC members are siblings of a brother or sister, with a disability or chronic health condition. There are different roles that each SibYAC member may choose to have with this study such as a listener, co-thinker, advisor or partner. The student researcher (LN) had a meeting with each SibYAC member and the whole SibYAC group using a conversation tool, the Involvement Matrix [21], to discuss the roles for how they would like to be involved in different phases of the study. The SibYAC have been involved in multiple aspects to design this study, including identifying the needs and goals of this study, developing the research question, identifying study methods, piloting the interview guide, and co-creating recruitment materials. Based on the experiences of SibYAC members, they identified that it is important to develop an understanding and raise awareness about the roles of siblings. They further suggested that there should be creative ways to engage with sibling participants. During the preparation of this study, they contributed to the co-development of recruitment materials (e.g., wording and language, visual appeal) to highlight the importance of participating in this study. Moving forward, discussions will be held bi-annually with the SibYAC using the Involvement Matrix about how they would like to continue to be involved in future stages of this study.

The lead student researcher (LN) was awarded a grant to provide compensation to SibYAC members. Currently, each SibYAC member is compensated for their time based on the guidelines outlined by the CHILD-BRIGHT Network [22]. The SibYAC members are also asked about how they would like to be compensated with different options.

Case study methodology

A descriptive single case study design will be used to understand the roles and responsibilities of siblings to support their brother or sister with a neurodisability in the preparation for health care transition [23]. This qualitative case study design is an empirical approach that investigates the case in-depth within a real-world context, when the phenomenon under study and the context in which it occurs cannot be delineated [24]. This study design was selected to understand the phenomenon of siblings' roles in supporting their brother or sister with a neurodisability during health care transition. Both the sibling who is typically developing and the sibling with a neurodisability have experiences as they are or were preparing for their roles during the developmental transition from adolescence to adulthood. Other types of case study designs have been applied to understand the experiences of youth with disabilities in life skills programs [25] and the social participation of young adults with autism spectrum disorders during the transition to adult life [26]. In this study, a single-case descriptive study design allows us to address "how" and "why" siblings took on certain roles to their brother or sister with a neurodisability.

Designing the case. A single case study can be conducted to describe circumstances and conditions of an everyday situation [23]. The definition of a case is necessary to identify the criteria for inclusion and exclusion of the participant sample. This study will define a single, holistic case as the experience of siblings with a brother or sister with a neurodisability preparing for health care transition. The following definitions will be provided to describe TD siblings as a study “case”: 1) sibling relationships and 2) childhood-onset neurodisabilities.

Sibling relationships. There are different types of families with varying definitions of sibling relationships. According to the American Academy of Pediatrics, there are three types of siblings: i) traditional siblings, in which brothers and sisters have the same mother and father (including those who are married, separated, or divorced); ii) half siblings who have the same mother or the same father; and iii) step siblings, in which brothers and sisters are not biologically related, but their parents are married to each other [27]. This study will adopt the holistic view of siblings as described by the American Academy of Pediatrics. Considering that sibling relationships can be unique, it is important to understand how siblings describe the factors involved in the roles and responsibilities they decided to take on when they have a brother or sister with neurodisability.

Childhood-onset neurodisabilities. A neurodisability is defined as a group of congenital or acquired long-term conditions due to an impairment of the brain and/or neuromuscular system that create functional limitations [28]. This study will focus on siblings with a childhood-onset neurodisability, which includes brain-based disabilities such as autism spectrum disorder, cerebral palsy, epilepsy, fetal alcohol spectrum disorder, and spina bifida [29].

Health care transition. Health care transition is defined as the “purposeful, planned movement of adolescents with chronic medical conditions from child-centred to adult-oriented health care” (10, p. 786). Health care transfer is an element of transition that is a one-time event when the youth transfer out of pediatric care to adult care [30]. Youth TD siblings who have a brother or sister with a neurodisability may have different roles to support this transition such as a mentor, role model, or friend [7].

Case binding

After defining a case, there should be boundaries placed on the case to ensure that the study is reasonable in scope, where there is a focused research question with clear study objectives [31]. An approach to bind the case is through geographic location and timeframe.

Geographical location. This study will focus on siblings living in Ontario. The geographical location of Ontario was selected to ensure that the case is representative of the common experiences of TD siblings with access to similar health care services.

Timeframe. Siblings who are typically developing will serve as a study “case” that is bound by their experiences during the developmental transition period of adolescence for both themselves and their sibling with a neurodisability [2]. Currently, there are few programs available for adolescent TD siblings with a brother or sister with a neurodisability which can affect the preparation of TD siblings in their roles as they age into adulthood.

Conceptual framework

A conceptual framework can be helpful to describe the propositions and possible relationships of constructs that will be identified in the study [31]. A conceptual framework will be developed as a starting point to describe possible factors that could influence siblings’ roles in supporting their brother or sister with a neurodisability. Based on a knowledge synthesis, a conceptual developmental framework was developed that outlined how interactions between a person and the environment can influence their relationships, resulting in certain outcomes [32].

Two theories provide further support for these constructs:

Transition theory [33]: Meleis and colleagues developed the transition theory to describe the different types of transition, the properties of the transition experience, and how individuals experience the process of transition resulting in specific outcomes [33]. Specific properties or characteristics of the sibling relationship, such as the emotions and attitudes of siblings towards each other as well as the level of closeness and frequency of contact between siblings [9, 34], can influence how siblings react to the trigger of health care transition and the outcomes that occurs regarding their roles and responsibilities.

Bioecological systems theory [35]: Urie Bronfenbrenner, a developmental psychologist, developed this theory to describe person-environment transactions, where the interaction between the youth with a neurodisability and multiple environments is a transactional process. This study will focus on the microsystem that encompasses individuals within the immediate environment of the TD sibling including parents, siblings, and health care providers as well as the mesosystem that includes the interactions between the microsystems such as the relationships with TD siblings, family members, and health care providers. Family roles could change when a youth with a neurodisability is preparing for health care transition [36]. There could also be changes in the sibling relationship, which is influenced by the context of the family that exists within the broader system of the external systems of health care, school, community, and policies. Figure 1 presents the initial conceptual framework, which will continue to evolve as the data are collected and analyzed. The final framework will include findings that address the propositions and constructs.

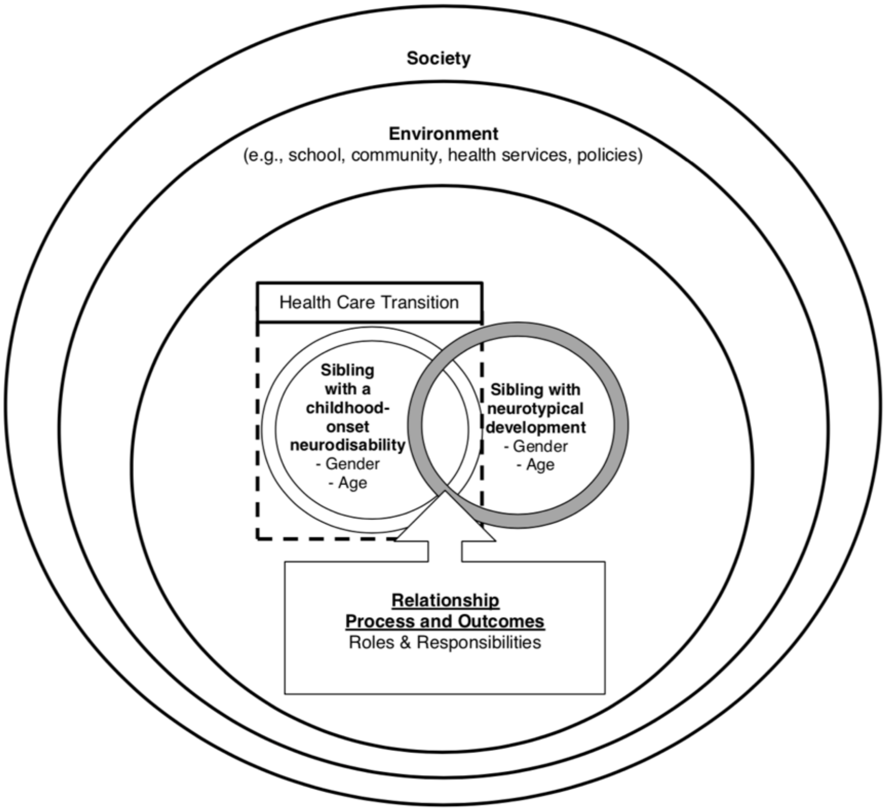

Figure 1. *Initial conceptual framework.*

Propositions

Propositions provide direction for factors that should be examined, which limits the scope of the study and increases the feasibility of completing the project [31, 37]. These propositions can serve as a guide for the study, such as data collection and analysis. Three propositions were developed based on the literature and the concepts that were included in the conceptual framework:

(1) *Gender.* Sibling dyads of the same gender are more likely to model one another, because they view similarities with each other [38]. We recognize that gender is non-binary and that siblings may identify themselves along a spectrum. In our study, we refer to siblings as a

"brother" or "sister" with the recognition that we will ask study participants about how they prefer to identify themselves.

(2) *Age.* Siblings who are older than their brother or sister with a neurodisability are likely to take on caregiving roles compared to younger siblings [7].

(3) *Health.* There may be differences in sibling roles depending on the overall health status and neurodisability diagnosis of their brother or sister [39].

Participant recruitment and sampling strategies

Participants will be recruited based on the following inclusion criteria: 1) between 14 and 40 years old; 2) able to speak English; and 3) has a brother or sister with a diagnosis of a childhood-onset neurodisability, such as autism spectrum disorder, cerebral palsy, Down syndrome, epilepsy, fetal alcohol spectrum disorder, or spina bifida between 14 and 21 years old. While adult siblings will be recruited in this study, they will be asked to reflect on their experiences during adolescence and how their roles and responsibilities to their brother or sister with a neurodisability might have changed over time. Participants will be recruited using social media, in which a website [40] has been created to share the study information. The recruitment materials include a study poster [41] and video that was co-created with the SibYAC [42]. These materials will be posted with a recruitment message on different social media platforms, such as Facebook, Twitter, and Instagram, and shared using the networks from the research team and reaching out to disability-specific organizations. A summary of how the recruitment materials have been shared is presented in Table 1. This study will also reach out to potential sibling participants of the brother or sister with a neurodisability from Ontario who is participating in an ongoing multi-regional Canadian randomized controlled trial [43]. In addition, snowball sampling will be used, in which participants in the study can refer other siblings [44]. Convenience and theoretical sampling will be conducted while data collection and ongoing data analysis is being conducted. Theoretical sampling allows for additional participants to be purposefully recruited who can provide in-depth rich information about the emerging codes, categories, and themes that may arise during data analysis [45]. Co-investigators, SibYAC members, and sibling participants will be asked to recruit potential siblings through their social networks. This study was approved by the Hamilton Integrated Research Ethics Board (REB Project 7932).

Table 1. A description of recruitment strategies.

Recruitment Strategy	**Description**
Social media postings	Recruitment materials with a key message about the importance of participating in this study will be shared on social media by co-investigators, SibYAC members, and by QUOI Media Group. Postings were made on different social media platforms, including Facebook, Twitter, and Instagram.
Organizations	Disability-specific and sibling-focused organizations, as well as child health networks can share the recruitment materials on their social media, including Facebook and Twitter. They can also share the recruitment materials in their monthly newsletters to staff, health care providers, and families. Organizations can also post the recruitment materials on their websites.
Posters and presentations	Presentations will be co-presented by LN with the SibYAC when possible, at local, national, and international conferences [for example, the CHILD-BRIGHT Network Symposium in May 2020, [46]]. Research posters will also be shared at conferences [for example, at the inaugural Canadian Transitions Pop-up Event [47]].
Social Networks	The recruitment materials will be shared by co-investigators, SibYAC members, and sibling participants to other siblings of individuals with a neurodisability who may be eligible to participate.

Sample size

The sample size for a case study methodology described by Yin (2018) depends on the number of participants that is sufficient to describe the phenomenon [23]. Qualitative studies conducted about TD siblings of a brother or sister with a disability have had a sample size of 10–20 participants [48]. This study aims to recruit approximately 20 participants with purposeful variation in age, gender, and the neurodisability diagnoses of their sibling.

Data collection

In this study, data will be collected through semi-structured, 1:1 in-depth interviews augmented by photo elicitation. Our SibYAC identified the novelty of photo elicitation to increase engagement with siblings as participants. They also believed that photo elicitation would enhance the dissemination of study findings with the potential to share photos through other avenues, such as a video to be posted on social media. Each participant will be invited to participate in a single interview of approximately 60–90 min. Due to public health measures and physical distancing guidelines from COVID-19 [49], all interviews will be conducted by phone or by videoconference (Zoom Communications Inc.) and participants may choose to turn on their web camera. During the interview, photo elicitation techniques will be used to deepen the nature of the discussion about their experiences with their sibling. Photo elicitation can be used with young adults to enhance their level of engagement in studies, especially since they are familiar with photography through their use of technology on social media, such as Instagram [50]. Photographs can help raise topics that may not necessarily be verbally be raised [51]. This study will use photo elicitation by asking participants to bring approximately 3–5 pre-existing photographs in preparation for the interview that exemplify their sibling experiences, including their roles and responsibilities related to supporting their brother or sister's preparation for health care transition. Electronic consent will be obtained from participants through Research Electronic Data Capture (REDCap™) [52], which is a secure web application to create and manage online forms (www.project-redcap.org) and will be hosted by the Department of Pediatrics at McMaster University. The consent process includes the following three steps [53]: i) consent to participate in the interview, including a description about their participation, benefits, and risks; ii) permission if participants share photographs from their parents or sibling; and iii) permission to share photographs on selected platforms (e.g., study publication, scientific presentations at conferences, workshops, videos, lay summaries, infographics). Steps two and three are optional, in which participants may choose to share photographs during the interview if they would like, but they can continue to participate in a verbal interview without sharing photographs. Participants can share the photographs before the interview by uploading them onto a secure cloud storage provided through McMaster's MacDrop (https://drop.mcmaster.ca/login). For all participants, the transcripts will be deanonymized to ensure confidentiality, and only the verbal descriptions of the photographs will be included in the analysis of the transcripts.

During the interview, each participant will also be asked to draw a family tree diagram, known as graphic elicitation of relational maps [54]. This type of technique has been previously used in research studies to actively engage with youth [54]. In this study, participants will be asked to draw on a piece of paper about the people in their immediate family. Guiding questions, which were modified from the demographic questions in the interview guide, will be used to encourage participants to draw the family tree diagram. After drawing the family tree diagram, participants can choose to show their drawing with their web camera or verbally describe their diagram. The process of drawing a family tree diagram will allow participants to begin to reflect on their thoughts, feelings, and experiences about their sibling relationships and overall family relationships [55]. Field notes will be written by LN after each interview, which will include both operational notes and analytic notes [56]. Operational notes will document information about the interview process, including questions to ask in subsequent interviews based on information that was raised in previous interviews, as the interview guide is semi-structured. Analytic notes will include information that addresses the research question about the roles and responsibilities of siblings of a brother or sister with a neurodisability.

Data management

All interviews will be audio-recorded, transcribed using Microsoft Word 2011, and imported into NVivo Version 11.4.2. All photographs will be scanned and imported into NVivo Version 11.4.2. All identifying information will be removed. All documents will be password protected.

Data analysis

The incorporation of graphic elicitation of relational maps with family tree diagrams and photo elicitation are techniques to provide an opportunity for participants to share in-depth details about their stories, and only the transcripts of the interviews will be analyzed. Reflexive thematic analysis, developed by Braun and Clarke [57], will be used to analyze the interviews with siblings. This analysis is comprised of six phases:

1. *Familiarizing oneself with the data.* The data will be read repeatedly, while taking notes and marking ideas for coding.

2. *Generating codes.* An initial list of ideas, representing codes, about what is interesting in the data will be generated. Analytic notes will also be reviewed to identify initial codes.
3. *Constructing themes.* The codes will be sorted into broader level themes. The relationship between codes, themes, and different levels of themes will be considered. The data will be visually represented, using tables and mind maps to help sort the codes into themes.
4. *Reviewing potential themes.* The themes will be reviewed to ensure that there is enough data to support them. Some themes may be reconsidered or collapsed into one theme.
5. *Defining and naming themes.* The themes will be further refined and defined to identify the key aspect that the theme captures. The scope and content for each theme will be described. The "story" will be identified to ensure that the data addresses the research questions.
6. *Producing the report.* The overall sibling's roles and responsibilities will be summarized as a report.

Discussion

A descriptive case study provides an opportunity to understand siblings' experiences, including their roles and responsibilities, when their brother or sister is preparing for the transition from pediatric to adult health care. One strength of this study is that there are multiple data sources, in which sibling participants can share photographs and family tree diagrams during the interviews. Sibling participants may feel empowered when they are provided with the opportunity to decide on the photographs that they wish to share and discuss in the interviews. They may also begin to think about the influence of their family relationships on their sibling relationship when they are drawing the family tree diagrams. This study is currently being conducted during the COVID-19 pandemic, and there may be novel findings about how the COVID-19 pandemic has impacted siblings' relationships. A recent study was published about the types of supports and worries that siblings of individuals with disabilities might have due to COVID-19 [58].

A second strength of this study is the use of social media as a recruitment strategy in order to adapt to physical distancing guidelines resulting from the COVID-19 pandemic. This study will document the different social media platforms that have been successful in reaching the audience of youth and young adult siblings who participated in this study. These recruitment strategies can inform future research even when physical distancing restrictions are relaxed.

A valuable aspect of this study is the ongoing collaboration with the SibYAC as research partners throughout the process of designing this study. The SibYAC have identified that this study is important to understanding and advocating for the roles of siblings when their brother or sister is preparing for health care transition. Findings from this study can help to tailor key messages to share with families about the involvement of siblings in health care, as well as to develop resources to support siblings' roles in health care.

A limitation of this study could be that participants are only recruited from Ontario, Canada, which could impact the generalizability of findings to other jurisdictions in Canada or countries. However, Ontario is a province that has the largest population size in Canada with almost 15 million people who live in a range of geographic locations, including urban and rural areas [59]. Based on the scarce literature, it is expected that this study will add significantly to the limited information on the roles and responsibilities of TD siblings of youth with a neurodisability with few resources to support TD siblings in their roles. This case study is designed to be bound by the geographical context of Ontario, and this geographical boundary allows for an in-depth analysis of the experiences of TD siblings who take on certain roles within a defined provincial health care, educational and social system. There may also be challenges with recruiting youth and young adult participants in this study. This study has implemented strategies to promote recruitment among young people, such as co-creating recruitment materials with the SibYAC where young people may want to hear from the perspectives of siblings about the importance of this study. The recruitment materials have been posted on social media, such as Facebook and Twitter, which are platforms that young people often engage with. This study also uses creative approaches to engage with participants during the study, in which participants can share photographs to describe their experiences as a sibling as well as draw a family tree diagram.

Conclusion

Siblings share a lifelong bond with their brother or sister, and siblings have a unique relationship in which they grow up together in the same home environment. When a brother or sister has a neurodisability, it is important to understand the different roles they choose to have in order to support siblings in these roles. This study aims to understand the roles and responsibilities of youth and young adult siblings who have a brother or sister with a neurodisability during the transition from pediatric to adult health care. An understanding of siblings' experiences along with best practices from the current literature can help to inform the development of resources or tools to support siblings in their roles in the health care of their brother or sister with a neurodisability.

Corresponding author: Linda Nguyen, PhD Candidate, School of Rehabilitation Science, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; and CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada, E-mail: nguyel7@mcmaster.ca

Funding source: Canadian Institutes of Health Research

Award Identifier / Grant number: Patient-Oriented Research Award - Transition to Leadership Stream - Phase 1

Funding source: CHILD-BRIGHT Network

Award Identifier / Grant number: Graduate Student Fellowship in Patient-Oriented Research

Funding source: Scotiabank Chair in Child Health Research

Acknowledgments

We gratefully acknowledge our partnership with the Sibling Youth Advisory Council throughout the design of this study.

Author contributions: All the authors have accepted responsibility for the entire content of the submitted manuscript and approved its submission.

Competing interests: Authors state no conflict of interest.

Research funding: This study was funded by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream held by LN. The partnership with the Sibling Youth Advisory Council was financially supported by the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network held by LN. Dr. Gorter holds the Scotiabank Chair in Child Health Research.

Informed consent: Not applicable.

Ethical approval: This study was approved by the Hamilton Integrated Research Ethics Board (REB Project 7932).

References

1. Stewart, D. Transition to adult services for young people with disabilities: current evidence to guide future research. *Dev Med Child Neurol* 2009;51(4 Suppl):169–73.

2. Sawyer, SM, Azzopardi, PS, Wickremarathne, D, Patton, GC. The age of adolescence. *Lancet Child Adolesc Health* 2018;2:223–8.

3. Freeman, M, Stewart, D, Cunningham, CE, Gorter, JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. *J Transit Med* 2018;1:20180003.

4. Blum, RW. Improving transition for adolescents with special health care needs from pediatric to adult-centered health care: introduction. *Pediatrics* 2002;110:1301–3.10.1542/peds.110.S3.1301

5. Stewart, DA, Law, MC, Rosenbaum, P, Willms, DG, Stewart, DA, Law, MC, et al.. A qualitative study of the transition to adulthood for youth with physical disabilities. *Phys Occup Ther Pediatr* 2009;21:3–21.

6. Whiteman, SD, McHale, SM, Soli, A. Theoretical perspectives on sibling relationships. *J Fam Theory Rev* 2011;3:124–39. https://doi.org/10.1111/j.1756-2589.2011.00087.x.

7. Hall, SA, Rossetti, Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. *J Appl Res Intellect Disabil* 2018;31:423–34.

8. Corsano, P, Musetti, A, Guidotti, L, Capelli, F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *J Intellect Dev Disabil* 2017;42:151–61.

9. Atkin, K, Tozer, R. Personalisation, family relationships and autism: conceptualising the role of adult siblings. *J Soc Work* 2014;14:225–42.

10. The Sibling Collaborative. Understanding the sibling experience: sibling needs assessment survey 2017 [Internet]; 2018. 1–19. Available from: http://cdn.agilitycms.com/partners-for-planning/Understanding the Sibling Experience–March 2018–Final.pdf [Accessed 2 Jun 2021].

11. Moyson, T, Roeyers, H. "The overall quality of my life as a sibling is all right, but of course, it could always be better". Quality of life of siblings of children with intellectual disability: the siblings' perspectives. *J Intellect Disabil Res* 2012;56:87–101.

12. Arnold, CK, Heller, T, Kramer, J. Support needs of siblings of people with developmental disabilities. *Intellect Dev Disabil* 2012;50:373–82.

13. McKenzie Smith, M, Pinto Pereira, S, Chan, L, Rose, C, Shafran, R. Impact of well-being interventions for siblings of children and young people with a chronic physical or mental

health condition: a systematic review and meta-analysis. *Clin Child Fam Psychol Rev* 2018;21:246–65.

14. Sibling Support Project. The sibling support project [Internet]; 2020. Available from: https://www.siblingsupport.org/ [Accessed 15 Nov 2020].

15. Opening Hearts. SibTeens [Internet]; 2019. Available from: https://www.openinghearts.ca/sibteens/ [Accessed 2 Jun 2021].

16. Craig, P, Dieppe, P, Macintyre, S, Michie, S, Nazareth, I, Petticrew, M, et al.. Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ* 2008;337:a1655.

17. Corsano, P, Musetti, A, Guidotti, L, Capelli, F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *J Intellect Dev Disabil* 2017;42:151–61.

18. Nguyen, T, Gorter, JW. Use of the international classification of functioning, disability and health as a framework for transition from paediatric to adult healthcare. *Child Care Health Dev* 2014;40:759–61.

19. Canadian Institutes of Health Research. Guide to knowledge translation planning at CIHR: integrated and end-of -grant approaches [Internet]; 2015. Available from: http://www.cihr-irsc.gc.ca/e/45321.html#a3 [Accessed 22 Apr 2019].

20. Richardson, SLL, Jordan, LS. Qualitative inquiry of sibling relationships: reinforcement of disability devaluation through the exclusion of voices. *Disabil Soc* 2017;32:1534–54.

21. Smits, D-W, van Meeteren, K, Klem, M, Alsem, M, Ketelaar, M. Designing a tool to support patient and public involvement in research projects: the involvement Matrix. *Res Involv Engagem* 2020;6:30.

22. CHILD-BRIGHT Network. Guidelines for patient partner compensation; 2020. Available from: https://www.child-bright.ca/compensation-guidelines [Accessed 23 Jun 2021].

23. Yin, RK. Designing case sudies. In: *Case study research and applications: design and methods*, 6th ed. Thousand Oaks, California: SAGE; 2018:25–80 pp.

24. Yin, RK. Getting started. In: *Case study research and applications: design and methods*, 6th ed. Thousand Oaks, California: SAGE; 2018:3–24 pp.

25. Kingsnorth, S, Rudzik, AEF, King, G, McPherson, AC. Residential immersive life skills programs for youth with disabilities: a case study of youth developmental trajectories of personal growth and caregiver perspectives. *BMC Pediatr* 2019;19:413.

26. Turcotte, P-L, Côté, C, Coulombe, K, Richard, M, Larivière, N, Couture, M. Social participation during transition to adult life among young adults with high-functioning autism spectrum disorders: experiences from an exploratory multiple case study. *Occup Ther Ment Health* 2015;31:234–52.

27. American Academy of Pediatrics. Sibling relationships [Internet]; 2007. Available from: https://www.ucy.ac.cy/nursery/documents/Sibling_Relation.pdf [Accessed 19 Apr 2019].

28. Morris, C, Janssens, A, Tomlinson, R, Williams, J, Logan, S. Towards a definition of neurodisability: a Delphi survey. *Dev Med Child Neurol* 2013;55:1103–8.

29. CHILD-BRIGHT Network. READYorNot(TM) brain-based disabilities project [Internet]; 2019. Available from: https://www.child-bright.ca [Accessed 15 Nov 2020].

30. Canadian Paediatric Society. Positon statement (AH 2007-01). Transition to adult care for youth with special health care needs. *Paediatr Child Health* 2007;12:785–8.

31. Baxter, P, Jack, S. Qualitative case study methodology: study design and implementation for novice researchers. *Qual Rep* 2008;13:544–59.10.46743/2160-3715/2008.1573

32. Gorter, JW, Stewart, D, Smith, MW, King, G, Wright, M, Nguyen, T, et al.. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. *Can J Community Ment Health* 2014;33:45–61.

33. Meleis, AI, Sawyer, LM, Im, EO, Messias, DK, Schumacher, K. Experiencing transitions: an emerging middle-range theory. *Adv Nurs Sci* 2000;23:12–28.

34. Davys, D, Mitchell, D, Haigh, C. Futures planning, parental expectations and sibling concern for people who have a learning disability. *J Intellect Disabil* 2010;14:167–83.

35. Bronfenbrenner, U. Six theories of child development: revised formulations and current issues. In: Vasta, R, editor. Ecological systems theory. London: Jessica Kingsley Publishers; 1992:187–249 pp.

36. Palisano, RJ, Di Rezze, B, Stewart, D, Rosenbaum, PL, Hlyva, O, Freeman, M, et al.. Life course health development of individuals with neurodevelopmental conditions. *Dev Med Child Neurol* 2017;59:470–6.

37. Yin, RK. Designing case studies: identifying your case(s) and establishing the logic of your case study. In: *Case study research and applications: design and methods*, 6th ed. Thousand Oaks, California: Sage Publications Inc.; 2018:25–78 pp.

38. Bandura, A. Toward a unifying theory of behavioral change. *Psychol Rev* 1977;84:191–215.

39. Orsmond, GI, Kuo, H-Y, Seltzer, MM. Siblings of individuals with an autism spectrum disorder. *Autism* 2009;13:59–80.

40. CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study [Internet]; 2021. Available from: https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study [Accessed 11 Apr 2021].

41. CanChild Centre for Childhood Disability Research. BEST SIBS study poster; 2021. Available from: https://canchild.ca/system/tenon/assets/attachments/000/003/528/original/Recruitment_Poster_(clean_copy)_-_February_8__2021.pdf [Accessed 11 Apr 2021].

42. CanChild Centre for Childhood Disability Research. BEST SIBS study recruitment video; 2021. Available from: https://vimeo.com/522169935/1e2b08984d [Accessed 11 Apr 2021].

43. Gorter, JW, Amaria, K, Kovacs, A, Rozenblum, R, Thabane, L, Galuppi, B, et al.. CHILD-BRIGHT READYorNot Brain-Based Disabilities Trial: protocol of a randomised controlled trial (RCT) investigating the effectiveness of a patient-facing e-health intervention designed to enhance healthcare transition readiness in youth. *BMJ Open* 2021;11:48756.

44. Patton, MQ. Designing qualitative studies. In: *Qualitative evaluation and research methods*, 2nd ed. Newbury Park: Sage Publications Inc.; 1990:145–98 pp.

45. Robinson, OC. Sampling in interview-based qualitative research: a theoretical and practical guide. *Qual Res Psychol* 2014;11:25–41.

46. Nguyen L, Davis H, Bellefeuille S, Ketelaar M, Di Rezze B, Jack S, Gorter JW, Sibling Youth Advisory Council. Sibling engagement in research: An illustrative example of a sibling-researcher partnership to design a study. CHILD-BRIGHT virtual symposium part 2. [Internet]; 2020. Available from: https://www.child-bright.ca/events/2020/4/29/symposium-pt2 [Accessed 11 Apr 2021].10.3389/fresc.2021.724589

47. Nguyen, L, Ketelaar, M, Di Rezze, B, Jack, S, Gorter, JW, Sibling Youth Advisory Council. The BEST SIBS study: a qualitative case study to understand the experiences of siblings of youth with a brain-based disability during health care transition [Internet]; 2021. Available from: https://www.childrenshealthcarecanada.ca/transitions-poster-hall/2021/1/11/the-best-sibs-study-a-qualitative-case-study-to-understand-the-experiences-of-siblings-of-youth-with-a-brain-based-disability-during-health-care-transition [Accessed 11 Apr 2021].

48. Avieli, H, Band-Winterstein, T, Araten Bergman, T. Sibling relationships over the life course: growing up with a disability. *Qual Health Res* 2019;29:1739–50.

49. Government of Canada. Coronavirus disease (COVID-19) [Internet]; 2020. Available from: https://www.canada.ca/en/public-health/services/diseases/coronavirus-disease-covid-19.html [Accessed 23 Jun 2021].

50. Sibeoni, J, Costa-Drolon, E, Poulmarc'h, L, Colin, S, Valentin, M, Pradère, J, et al.. Photo-elicitation with adolescents in qualitative research: an example of its use in exploring family interactions in adolescent psychiatry. *Child Adolesc Psychiatr Ment Health* 2017;11:1–11.

51. Croghan, R, Griffin, C, Hunter, J, Phoenix, A. Young people's constructions of self: notes on the use and analysis of the photo-elicitation methods. *Int J Soc Res Methodol* 2008;11:345–56.

52. Harris, PA, Taylor, R, Thielke, R, Payne, J, Gonzalez, N, Conde, JG. Research electronic data capture (REDCap)—a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inf* 2009;42:377–81.

53. Wang, CC, Redwood-Jones, YA. Photovoice ethics: perspectives from flint photovoice. *Health Educ Behav* 2001;28:560-72.

54. Bagnoli, A. Beyond the standard interview: the use of graphic elicitation and arts-based methods. *Qual Res* 2009;9:547–70.

55. Kearney, KS, Hyle, AE. Drawing out emotions: the use of participant-produced drawings in qualitative inquiry. *Qual Res* 2004;4:361–82.

56. Birks, M, Francis, K. Memoing in qualitative research probing data and processes. *J Res Nurs* 2008;13:68–75.

57. Braun, V, Clarke, V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.

58. Redquest, BK, Tint, A, Ries, H, Lunsky, Y. Exploring the experiences of siblings of adults with intellectual/developmental disabilities during the COVID-19 pandemic. *J Intellect Disabil Res* 2021;65:1–10.

59. Statistics Canada. Population estimates, quarterly [Internet]; 2021. Available from: https://www150.statcan.gc.ca/t1/tbl1/en/tv.action?pid=1710000901 [Accessed 11 Apr 2021].

CHAPTER SIX

TITLE: Roles and responsibilities of siblings of youth with a neurodisability during the transition to adulthood: A qualitative case study

AUTHORS: Nguyen L, Jack SM, Davis H, Bellefeuille S, Arafeh D, Di Rezze B, Ketelaar M, & Gorter JW

JOURNAL: To be prepared for submission to a peer-reviewed journal focused on the health and development of children, adolescents, and young adults.

Abstract

Background: During the transition to adulthood, youth with a neurodisability may face challenges including learning how to navigate services in the adult care system. Youth can turn to their families, including siblings, for support. However, the sibling roles and responsibilities during this transiton period are unclear. This study aims to understand the roles and responsibilities that siblings have in their relationship with their sibling with a neurodisability.

Methods: This study uses a descriptive single case study. Siblings were eligible to participate if they were between 14 to 40 years old, had a sibling between 14 to 21 years with a childhood-onset neurodisability, and spoke English. Semi-structured interviews augmented by techniques of photo elicitation and relational maps were analyzed using reflexive thematic analysis. Our team partnered with siblings with lived experience who informed all study phases.

Results: Nineteen participants (mean age of 19.5 years, standard deviation of 5.0 years) from 16 unique families were interviewed. Six unique roles were described including friend, role model/mentor, protector, advocate, supporter or caregiver. Multiple levels of emotional responsibility were associated with each role over the life course trajectory: low level for friend during childhood; medium level for model/mentor, protector, advocate or supporter during adolescence; and high level for caregiver during emerging adulthood. Siblings identified the level of emotional responsibility based on the amount of processing required to balance their personal lives with the well-being of their sibling with a neurodisability. Intrapersonal characteristics, including personal identity, values, and experiences, influenced roles assumed by siblings. Siblings also described a four-phase decision-making process for their roles: 1) acquiring knowledge; 2) preparing plans; 3) making adjustments; and 4) seeking supports.

Conclusions: Findings highlight that siblings require supports as they process their decisions and emotional responsibility in their roles. Resources can be developed or further enhanced to support siblings.

Keywords: healthcare transition, youth, disabilities, siblings, roles, qualitative

Key Messages

1. Partnering with siblings with lived experiences was valuable to highlight the need of this study, deepen the level of understanding and interpretation of findings, and discuss the implications of the findings.
2. Siblings assume multiple roles over the life course trajectory based on a distinct decision-making process.
3. Each sibling role is associated with a level of emotional responsibility that balances the personal interests of the sibling with the well-being of the sibling with a neurodisability.
4. There is a critical need to provide supports for siblings of individuals with a neurodisability as they transition into their roles.

Introduction

Youth transitioning to adulthood typically explore their interests and goals related to different opportunities for education, employment, leisure, and relationships (1). These interests are often explored during the developmental periods of adolescence and emerging adulthood, during the ages of 12 to 25 years (2). Youth with a neurodisability, such as autism spectrum disorder or cerebral palsy, often face additional challenges during these developmental stages and in particular during healthcare transition when they need to learn how to navigate services in the adult healthcare system (3). Healthcare transition is defined as the “purposeful, planned movement of adolescents with chronic medical conditions from child-centred to adult-oriented healthcare” (4). Youth and their families, including siblings when present, need to be prepared for healthcare transition.

Siblings have an important role in the family, but their role during the transition phase from adolescence to adulthood, and during healthcare transition is often unclear. Throughout childhood, typically developing (TD) siblings can have different roles to their sibling with a neurodisability such as a mentor, role model, or friend (5). Siblings can spend proportionately more time with their sibling with a neurodisability compared to any other family member, and may have multiple roles (6).

Despite the unique relationship between siblings, many families may not have or may delay formal discussions about the roles and expectations of siblings until after the process of transition (6). This can lead to missed opportunities to be engaged in healthcare and increased anxiety about future roles for TD siblings. Siblings of individuals with a disability have expressed their concerns about their present and future care responsibilities. In Canada, a needs assessment survey was conducted in 2018 among siblings ages 20 years and older who had a sibling with an intellectual and/or developmental disability (7). Within the sample, approximately 71% of siblings expected that they would provide increasing caregiving support to their sibling with a disability in the future (7). Siblings often have worries about responsibilities in the future, such as guardianship or financial responsibilities that they might take on when their parents can no longer be the primary caregivers (8). Siblings may process these different concerns and consider the extent to which they choose to have certain roles to their siblings with a neurodisability.

Siblings have held different types of responsibilities to their sibling with a disability. A review and analysis of online resources found that siblings shared in blogs and interviews about the roles and responsibilities that they have to their sibling with a neurodisability, such as providing support and attending therapy appointments (9). Siblings also shared about their emotions associated with roles, such as frustration, guilt, stress, but also love and care for their sibling with a neurodisability (9). While siblings have shared brief descriptions of their roles, details are needed about the types of roles that siblings have to their sibling with a neurodisability during healthcare transition. An in-depth understanding about these roles from the perspectives of siblings with lived experiences can help identify the types of resources to support them in their roles.

Objectives

This BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study aims to: 1) describe sibling roles, including any functions associated with healthcare transitions; and 2) identify the siblings’ responsibilities in their relationship with their brother or sister with a neurodisability.

Methods

Engagement of Siblings with Lived Experience in the Research Process

Patient-oriented research is an approach to engage patients throughout all phases of research (10). Patients can be an overarching term that includes individuals with experience of a health issue, as well as their informal caregivers including family and friends (10)The engagement of patients and their family, including siblings, with lived experience is important to ensure that their voices are represented in research. The incorporation of these experiences can help enhance the value and relevance of the research is relevant and provide opportunities to share the study results with target audiences (11–13). To accomplish this goal, the Sibling Youth Advisory Council (SibYAC) was established in 2018 by the first author (LN) as a research partner. The SibYAC includes six young adult siblings, with five sisters and one brother, between 21 to 28 years old, of youth with a disability. To ensure that the partnership was meaningful and reflected the lived experiences of the SibYAC, key principles for engagement with patients and families were applied such as creating an inclusive environment to integrate lived experiences into the research, providing support and flexibility for the SibYAC to contribute to the research; ensuring mutual respect by valuing the expertise and knowledge from the SibYAC, and providing opportunities to co-build the research together (12,14,15).

Throughout the study, conversation tools, including the Involvement Matrix (16) and Engagement Tool (17), were used to guide ongoing discussions with each SibYAC member to understand how they would like to be engaged in each study phase. Since the inception of the study, members of the SibYAC have actively engaged in multiple study phases. These phases included: 1) preparation, such as co-creating recruitment materials and pilot testing the interview guide; 2) execution, such as recruiting participants by sharing the study information in their networks; and 3) knowledge implementation, such as co-presenting experiences of our partnership at local, national, and international conferences.

Case Study

This descriptive single case study qualitatively explored the roles and responsibilities identified by siblings of a sibling with a neurodisability during healthcare transition. A detailed description of the study methods has been published (16).

Study Participant Recruitment

To describe the unique roles and responsibilities of siblings, a purposeful sample of individuals living in families where they were a sibling to another individual with a disability were recruited. Siblings were eligible to participate if they: 1) were between 14 to 40 years old; 2) have a sibling between 14 to 21 years old with a childhood-onset neurodisability; and 3) spoke English. The SibYAC were involved with co-creating recruitment materials including a poster (17), video (18), and website (19) that were shared on social media platforms (e.g., Facebook, Twitter), the personal networks of the research team and disability-specific organizations such as the CHILD-BRIGHT Network (20) and Kids Brain Health Network (21). Study information was also shared with the siblings of a sibling with a neurodisability who participated in a randomized controlled trial (22). Snowball sampling was also used in which study participants could share the study information with other potential siblings who may be interested in participating in this study (23). Participants were recruited from February 2020 to July 2021. Data generation and data analysis occurred concurrently. Participant recruitment was completed when there was a complete description with the dimensions and properties of the identified roles that siblings had to their sibling with a neurodisability. In this paper, the term ‘sibling participant’ will be used instead of ‘TD sibling’ as a participant was not excluded if they had a neurodisability.

Ethical Approval

This study received ethical approval from the Hamilton Integrated Research Ethics Board (Project #7932). Consent was obtained in a three-step process (24): 1) to participate in the interview; 2) to share photographs, including those from their parents and/or sibling; and 3) to share photographs on specific platforms (e.g., study publication, presentations at conferences). Steps two and three were optional, in which participants could choose to participate in the interview without sharing photographs. At the beginning of each interview, information about confidentiality and privacy was reviewed with participants. Participants could choose to not answer specific questions or stop the interview at any time. They were also asked if there were any measures that could be considered during the interview in order to feel more comfortable. This information was important to share as all interviews were conducted via a web platform.

Data Collection

The triangulation of data sources is a hallmark characteristics of case study research. In this study, a comprehensive description of the case was established through generating data from three sources: interviews, photo elicitation, and graphic elicitation of relational maps. Semi-structured, one-on-one single interviews scheduled for approximately an hour were conducted with participants to explore their roles and responsibilities to their sibling with a neurodisability.

These interviews were conducted by the first author (LN), and because of public health measures to reduce the transmission of SARS-Cov-2, were conducted via a web conference platform. To enhance rapport and increase the quality and detail of information shared, two elicitation techniques augmented the in-depth interviews. Using photo elicitation, participants were asked to share 3-5 photographs that described their sibling experiences, which could be brought to the interview or uploaded before the interview on a secure cloud storage. Then to understand the family structure, and the relationships between family members, graphic elicitation of relational maps was used where participants were asked to draw a family tree diagram of members in their immediate family to discuss the context of their family dynamics and their role within the family. A summary of the questions for the interviews are provided in Table 1.

Table 1. Summary of interview questions.

Questions asked during the graphic elicitation of relational maps 1. To start, can you draw/write about who you are? 2. Who lives at home with you? 3. Are there any family members who do not live at home with you, but you also often talk with or ask questions? 4. Can you draw/write who your siblings are? 5. How old is each of your siblings? 6. What grade is each of your sibling in? 7. Which sibling has a disability? What type of disability does your sibling have?
Questions asked during photo elicitation 1. Can you tell me about this picture? 2. Who is in the picture? 3. Where was this picture taken? 4. Why did you take this picture? 5. What does this picture mean to you? 6. Is there a picture that you wish that you had?
Questions asked to describe sibling relationships 1. How would you describe your sibling relationship? 2. One of your siblings is currently preparing for health care transition. When your sibling turns 18 years old, they will have to transfer from pediatric to adult care. Have you heard about this before? 3. As you grow up, how do you see the future of you and your siblings as adults?

4. As a sibling to another sibling with a disability in your family, what is the most important information about your personal experiences would be most important for other people, including health care professionals to understand?

Data Analysis

Data generation and data analysis occurred in an iterative process throughout participant recruitment. Interview transcripts, including the verbal descriptions of the photographs and family tree diagrams, were analyzed using reflexive thematic analysis developed by Braun and Clarke (25). The first author (LN) wrote field notes after each interview to identify initial codes and categories. These categories were reviewed and discussed among stakeholders from a range of educational backgrounds in Canada, including cognitive psychology, education, nursing, occupational therapy, physiatry, rehabilitation, user experience design, patient-oriented research, and lived experiences. The stakeholders included the SibYAC and five graduate students who were purposefully identified based on their diverse educational backgrounds and were connected with this research team. Stakeholder consultation was a form of analyst triangulation and peer debriefing to enhance the credibility of the data (26).

Conceptual Framework

The application of a conceptual framework is necessary in a case study and congruent with reflexive thematic analysis (27,28). The use of a conceptual framework ensured that there were boundaries about the case of sibling participants with a clear focus for data analysis (27). A conceptual framework was developed to guide the initial step of data analysis using two theories:

1. *Transition theory* (29)*:* Meleis and colleagues developed this theory to describe the different types of transitions, properties of transition, and outcomes experienced by individuals during the process of transition.

2. *Bioecological systems theory* (30): Urie Bronfrenbrenner developed this theory to describe person-environment transactions. The application of this theory helped to identify the focus on the microsystem comprised of individuals in the immediate family environment of the TD sibling including the sibling with a neurodisability, siblings, and parents.

The propositions included in the initial conceptual framework provided guidance about the factors to analyze that could influence the relationship and roles that sibling participants have with their sibling with a neurodisability. The propositions analyzed included gender, age, and health of the sibling participant and sibling with a neurodisability.

Results

Nineteen sibling participants with a mean age of 19.5 years (standard deviation [SD] = 5.0) from 16 unique families were included. Fourteen participants described their relationship to one sibling with a neurodisability and five participants described their relationship to two siblings with a neurodisability, for a total of twenty unique siblings with a neurodisability (mean age 16.9 years, SD = 2.1) during the interviews. Three sibling participants disclosed that they had a neurodisability. Table 2 describes the characteristics of the siblings and families. Interviews with participants ranged from 45 to 104 minutes.

Table 2. Demographics of sibling participants.

Characteristics	**Total number**
Gender of sibling participants	
Female	12
Male	7
Gender of sibling with a neurodisability	
Female	5
Male	15
Type of neurodisability of the sibling	
Autism spectrum disorder	12

Cerebral palsy	4
Hereditary spastic paraplegia	1
Intellectual disorder	1
Attention deficit hyperactivity disorder	1
Epilepsy	1
Sibling dyad (sibling participant-sibling with a neurodisability)	
Sister-brother	11
Sister-sister	3
Brother-sister	4
Brother-brother	6
Birth order (sibling participants to their sibling with a neurodisability)	
Older sister, brother	7
Older sister, sister	3
Older brother, sister	4
Older brother, brother	2
Younger sister, brother	4
Younger sister, sister	0
Younger brother, sister	0
Younger brother, brother	4
Number of siblings with a neurodisability in the family	
1 sibling with a neurodisability	15
2+ siblings with a neurodisability	4
Total number of siblings in the family	
2 siblings	8
3 siblings	6
4+ siblings	5

Roles

Sibling participants described six unique roles to their sibling with a neurodisability, which included friend, role model/mentor, protector, advocate, supporter, or caregiver (Figure 1). The type of sibling role differed across the life course trajectory and the degree of emotional responsibility invested by the sibling in their roles varied. Detailed descriptions about each role are provided in Table 3.

[Insert Table 3 about here]

Different levels of emotional responsibility were associated with each role as seen in Figure 1, categorized as “low”, “medium”, or “high”. Emotional responsibility is defined as the accountability that an individual has for specific emotions (31,32). While individuals are responsible for the emotions that they feel, these emotions represent their state of mind (32). Sibling participants described how they learned to control these emotions in order to best assume their role to their sibling with a neurodisability. During a sibling's life course, one of the first roles assumed was that of a friend. Siblings described playing with their sibling with a neurodisability as having a low level of emotional responsibility. They could simply play with their sibling with a neurodisability as they would with their other siblings or peers.

During adolescence, sibling participants began to take on the roles of being a model/mentor, protector, advocate, or supporter, which carried a medium level of emotional responsibility. Siblings felt that they had to be present for their sibling with a neurodisability by offering guidance, yet they also wanted to prevent their sibling with a neurodisability from feeling negative emotions. In the role of a protector or advocate, for example, sibling participants described situations where they remained positive in front of their sibling with a neurodisability, but they also had to explain the neurodisability to others who might not understand such as their peers or other family members.

As sibling participants entered emerging adulthood, some described that they were either preparing for or had the role of a caregiver. This role had the highest level of emotional responsibility, because sibling participants needed to ensure that the basic needs of their sibling with a neurodisability were met such as assistance with therapy and personal hygiene. Some sibling participants gradually took on these caregiving tasks that was implicitly expected from the family, whereas others actively chose to have these tasks. Sibling participants felt multiple emotions, such as guilt, as they were processing how they could continue with caregiving tasks while balancing their own needs such as employment and/or post-secondary education. The dynamic balance of emotions in the role of a caregiver carried a high level of emotional responsibility, as sibling participants had to maintain control of their emotions.

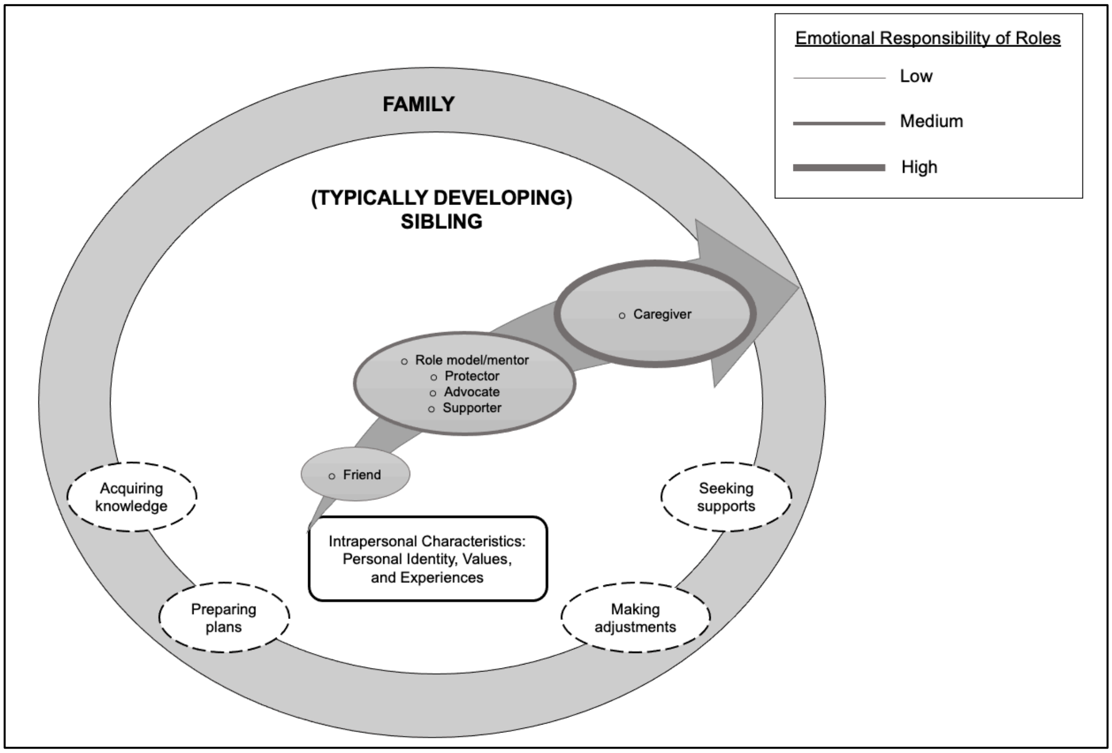

Figure 1. *The internal process of factors that influenced the roles that sibling participants had to their sibling with a neurodisability.*

Intrapersonal characteristics

Siblings perceived that certain intrapersonal characteristics, including their personal identity, values, and experiences, influenced their adoption of different roles.

Personal identity

Personal identity was comprised of their gender and age, including the birth order to their sibling with a neurodisability. Many sibling participants older than the sibling with a neurodisability were often a caregiver, role model, or mentor to their sibling with a neurodisability. Older sibling participants also described that they wanted to take on the role of being a protector, for example, to physically protect their sibling with a neurodisability from bullying. Some sibling participants wanted to protect the sibling with a neurodisability from feeling negative emotions by not oversharing their own personal conflicts and stresses.

Gender further influenced the types of roles that sibling participants had. Twelve sibling participants identified as females, and many of them chose or were expected to have caregiving responsibilities. For some siblings, there was an implicit expectation from parents that the sibling participants would assist with activities of daily living, such as personal hygiene or preparing meals, for the sibling with a neurodisability. While sibling participants did not identify a specific age for when they took on certain roles, they described how they naturally began to take on more responsibilities to their sibling with a neurodisability. Initially, they stayed at home with their sibling with a neurodisability when their parents were not at home. They slowly began to take on more tasks such as picking up their sibling with a neurodisability and provide support with therapy. Many sisters felt the need to become guardians to their sibling with a neurodisability regardless of their age, as one sister describes:

> I have been starting to think about it [the future]. My parents are gonna get old and then I might be my own and I know that I have to take over what they were doing, so booking all their appointments and understanding what their needs are. I know that it's going to be up to me to make sure that they're getting everything that they [brothers with autism spectrum disorder] need" (Younger sister of brothers with autism spectrum disorder).

Sibling participants, especially sisters, often thought about the future and how they could provide caregiving support to their siblings with a neurodisability. Most brothers also cared for their siblings with a neurodisability, but they were often a friend or supporter such as comforting their sibling with a neurodisability during family conflicts.

Value of familial responsibility

The value of familial responsibility was identified by many sibling participants as being important, which influenced their roles. For example, an older sister describes how her value of familial responsibility were instilled by her parents:

> So it's like they've always encouraged us to kind of bond with each other, like spend time with each other. Even with me going off into university, like making sure that we still remain in contact because... as my parents grow older, I don't know when they're going to move on so... it has to be like me and my brother kind of navigating life together. (Older sister of a brother with autism spectrum disorder).

Although some sibling participants did not mention how they came to adopt this family value, many sibling participants felt this familial responsibility, in which they were expected to take care of their sibling with a neurodisability.

Experiences

Among some sibling participants, their values were influenced by personal experiences. Some sibling participants shared that their own experience of having the same neurodisability as their sibling meant that they could understand the challenges that their sibling was experiencing. For example, some sibling participants did not have a good experience with certain medications or tried different types of therapy. Based on these personal reflections, sibling participants wanted to share their experiences about how they overcame certain challenges to manage their neurodisability with their sibling with a neurodisability. They had the value of empathy, in which they wanted to make sure that they could support their sibling with a neurodisability through similar challenges that they faced and overcame. Overall, sibling participants who shared experiences of having the same neurodisability as their sibling perceived that they were in a stronger position to be a mentor and offer guidance.

Decision-making process

As siblings reflected on their experiences as both they and their sibling aged, there was a distinct decision-making process describing how siblings determined which roles and responsibilities they would assume. This four-phase process consisted of: 1) acquiring knowledge; 2) preparing plans; 3) making adjustments; and 4) seeking supports.

Acquiring knowledge. Sibling participants identified that they needed to acquire knowledge about how they can best support their sibling with a disability. Often, they turned to their parents for knowledge about the disability. When they first learned about the diagnosis, which was usually through conversations with their parents, they had questions about the diagnosis and what the diagnosis meant for their roles as siblings. The types of questions that sibling participants asked depended on their age. If they learned about the diagnosis as young children, they required explanations about what the diagnosis looked like. In some families, the siblings received their diagnosis of having a neurodisability during adolescence. Siblings who were adolescents or young adults had more specific questions about how the diagnosis of their siblings might affect their life. An older brother described the questions that he asked when he first learned about the diagnosis of his sister:

> How will my day-to-day life need to change so that I can like help and make sure things go smoothly with her? I think those were probably my first questions.... 'What just happened?'... 'What will we need to do now just to make it easier for everyone and... just a smooth transition to her new way of life' (Older brother of a sister with epilepsy).

For all sibling participants, they wanted to have sufficient knowledge about how they can best support their sibling with a neurodisability.

Preparing plans. Sibling participants described how they needed to prepare plans, which included how they coordinated plans with other siblings and members in the family. For example, they needed to make plans with their friends and family if they had to stay at home to take care of their sibling with a neurodisability. A sister described her feelings when she was not able to socialize with her friends due to her responsibility with her brother with cerebral palsy:

> They [friends] will invite me out... but you know, sometimes I'll have to refuse because I can't be away from home that day... I think it's hard for them to understand no matter how hard I try to explain it to them that my role at home is important. (Older sister of a brother with cerebral palsy).

Families needed to prepare for the transition to adulthood of the sibling with a neurodisability, and parents were often focused on two transitions: the transition from pediatric to adult healthcare and the transition after graduating from high school. Although sibling participants were not actively involved in preparing transition plans, they were kept informed about steps that were being taken to help the sibling with a neurodisability prepare for transition. For example, they were aware about the resources that were being accessed by the family.

Future plans also included steps of what might happen when their parents passed away. Some families discussed that the sibling participants were not expected to become the official guardians, but they would still have a prominent role in deciding who the official guardians might be. For other sibling participants, guardianship was not openly discussed but they thought about the possibility that they might become the official guardians.

Making adjustments. Sibling participants were constantly reflecting and making adjustments to their short-term and long-term plans. When their sibling with a neurodisability first received a diagnosis, some sibling participants made adjustments in which they asked their parents if they could provide support. For example, picking up or being at home with their sibling with a neurodisability. Sometimes these adjustments in plans required negotiation between siblings and family members, such as whether someone else could take on responsibilities for the day. A sister describes how she coordinates responsibilities with her family:

> There are so many things that we need to do to make sure [sibling with cerebral palsy] gets the best care. It's just kind of a lot of communicating.... Or dividing up the roles and what needs to be done, among me, my sister, and my parents. (Older sister of a brother with cerebral palsy).

In many families, there was ongoing communication about adjustments to plans especially during the COVID-19 pandemic. When interviews were conducted with sibling participants, public health guidelines included physical distancing and virtual online learning. Sibling participants reflected on how their relationship with their sibling with a neurodisability and family changed during the COVID-19 pandemic. There was a challenge that sibling participants faced, in which they began to think about how to communicate with their sibling with a neurodisability during the transition out of lockdowns. Many siblings with a neurodisability struggled with the lack of routine at home. Families had to communicate that the lockdowns were temporary and the siblings with a neurodisability would have an opportunity to return to school in-person soon. Sibling participants realized that when lockdown restrictions were lifted, they would likely move away from home again and they needed to prepare their sibling with a neurodisability for this change. In many families, conversations were already taking place with the sibling with a neurodisability prior to the COVID-19 pandemic but the pandemic provided some more time for the siblings with a neurodisability to adjust to plans.

Seeking supports. All sibling participants discussed seeking supports for their mental health and well-being. Sibling participants experienced a spectrum of emotions. It was common across participants to experience feelings of guilt when they had negative reactions (e.g., frustration, sadness, annoyance) towards their sibling with a neurodisability. An older sister describes the guilt that she feels when she spends time with friends:

> Even though there's a sense of, in the moment when I'm refusing to go out 'I wish I can go' and the presence of guilt almost that I can't go, but when I come out and see [name of sibling with cerebral palsy], yeah, this is what I'm doing it for. And then even when I am

> out with my friends... there's a sense of guilt... because I keep thinking in my head, 'Like okay, I should be at home, I wonder if [name of sibling with cerebral palsy] is okay (Older sister of a brother with cerebral palsy).

Sibling participants sought different resources to manage and cope with their emotions. During childhood, sibling participants often shared their emotions with their parents. They frequently turned to their mothers for emotional support who could provide comfort and information to explain the behaviour of the sibling with a neurodisability. As sibling participants grew up, they turned to their older siblings for advice and support. For sibling participants who did not have other siblings in the family, they often independently sought therapy from healthcare professionals. Some sibling participants felt that they could connect with friends who were also siblings of individuals with a neurodisability, because they had similar experiences and could share advice. Support was critical for all sibling participants to cope with the array of emotions that they felt as a sibling of an individual with a disability.

Discussion

This study identified how sibling participants had different roles that evolved over the life course trajectory, especially during the time when both the sibling participants and their sibling with a neurodisability were transitioning to adulthood. Similar roles have been identified by a study conducted by Hall and Rossetti (2017), in which an online survey was shared with adult siblings of individuals with intellectual and developmental disabilities (5). This study further builds on existing literature in which intrapersonal characteristics, including their personal identity, values, and experiences, influenced the roles that sibling participants had to their sibling with a neurodisability. Other studies identified similar variables that influenced how TD siblings chose to have the specific role as a caregiver (33–35). Adult siblings were expected to provide future care if they had no other siblings, identified as female, lived close in proximity, and had a close emotional relationship with their sibling with an intellectual or developmental disability (33,34). Many sibling participants in this study who identified as female and were the only typically developing sibling in the family were already a caregiver or were expected to have this role in the future.

This study also identified that siblings had a distinct four-step process in how they decided on their roles: acquiring knowledge, preparing plans, making adjustments and seeking supports. Previous studies focused on the perspectives of adult siblings about their plans with caregiving to their sibling with a disability (34,36,37). Siblings are in a unique position as the "sandwich" in which they are considering how they might care for their sibling with a neurodisability, and also their aging parents (38,39). Often, there are few discussions in the family about future planning (40–42). Some families, similar to the experiences of sibling participants, may choose not to have discussions about future plans because the event for changes in roles appears to be in the far-off future (33). The perspectives of sibling participants in this study further adds to the current literature in that families might not have discussions about future planning, but sibling participants as young as age 14 were already having these thoughts. Despite the need for discussions about future planning with the whole family, our previous review of the grey literature about resources for siblings to support with healthcare management identified no resources to facilitate family conversations for future planning (9).

Sibling participants described additional barriers that their families faced in systems, such as the educational and health system, when their sibling with a neurodisability was transitioning to adulthood. Their parents were actively seeking for programs and resources to support multiple transitions that their sibling with a neurodisability was experiencing. Similar findings were identified in a systematic review, in which ten studies reported the challenges that siblings of individuals with an intellectual or developmental disability faced with navigating systems (34). Current and future sibling caregivers described that there is little information or guidance about how to navigate different services (34). Members of the SibYAC described the valuable role that they have to their sibling with a disability, and their roles needed to be acknowledged by parents, service providers and professionals (43).

Supports are also required for the emotional health and well-being of siblings, as described by sibling participants and the SibYAC (43). All sibling participants received supports from different sources, such as their peers with or without a sibling with a neurodisability, parents, or healthcare professionals. In Canada, siblings of individuals with a neurodisability can receive supports in different ways, such as connecting with support groups; for example in Canada, there are Sibshops for ages 8-13 years old (44), SibTeen sessions for ages 13-17 years old (45), and the Sibling Collaborative for adult siblings (46).

Strengths and Limitations

A strength of this study is the engagement of multiple stakeholders, including our multidisciplinary team and SibYAC, which were critical to the elucidation of key themes that captured the multi-layered factors that influenced how TD siblings chose different roles over time. A second strength was the use of an initial conceptual framework supported by the transitions theory (29) and bioecological systems theory (30) to focus the scope of this study on the roles and responsibilities described by sibling participants.

A limitation of this study was that most sibling participants were older females who may have more interest in sharing their roles to their sibling with a neurodisability compared to siblings who did not participate. Based on the literature, female siblings often plan to become future caregivers to their sibling with a neurodisability (33,34). However, this study also allowed for a deep understanding of how siblings processed their decision to become caregivers. Another limitation is that multiple family members such as the youth with a neurodisability and parents/caregivers could have been interviewed to understand their perspectives about the roles of TD siblings in the family. This study only focused on the perspectives of siblings about their roles.

Future Directions

This study is part of a doctoral research program (LN). As part of this program, two reviews have been conducted including a scoping review of the published literature about the characteristics of programs (47) and a second review and qualitative document analysis of grey literature resources available on websites of organizations that are part of Children's Healthcare Canada (9). This study described the perspectives of siblings and identified a need develop resources to support siblings in their roles. A next step is to co-develop resources with multidisciplinary stakeholders that could potentially help siblings in their roles who can then support their sibling with a neurodisability during the transition to adulthood.

A second step is to conduct further research about the intersection between personal, cultural, and family values that can influence the roles of siblings of youth with a neurodisability. This study provided preliminary information about how these values affect the initial step to the selection of sibling roles. For example, certain cultures emphasize familial responsibility in which siblings may feel an obligation to have a caregiver role to their sibling with a neurodisability (35,48). Sibling participants also identified that they had personal values and so, a further understanding is needed about how different values are connected and formed to influence sibling roles.

A third step is to ensure that there are future supports provided for siblings during emerging adulthood. This developmental period of emerging adulthood is a critical time period where the person and environment is changing, and a positive support system can optimize the growth of the person (49). There remains a gap that has been identified by the SibYAC, in which there are limited support groups for young adult siblings ages 17 to 25 years old. Members of the SibYAC reflected on the personal value of being a part of this advisory council, which has become a support network for them.

Conclusion

Since childhood, siblings are navigating roles and responsibilities to their sibling with a neurodisability. These roles continue to change during a time when both sibling participants and their sibling with a neurodisability were transitioning to adulthood. Each role comes with its own emotional responsibility that siblings have, yet they all want to be present to provide the best support and care that they can to their sibling with a neurodisability. Future resources are needed to prepare and support siblings as they transition to their roles.

Acknowledgements

We are grateful to the siblings who participate in this study. We gratefully acknowledge our partnership with the Sibling Youth Advisory Council (SibYAC) throughout all study phases, members were asked about their level of involvement and chose whether they wanted to have a co-authorship role. We also acknowledge our colleagues involved in the peer debriefing process of the analysis phase: Carrie Bullard, Shelby Deibert, Tessa Elliott, Paige Gehrke, and Carly Whitmore.

Disclosure statement

The authors have no conflicts of interest.

Funding

This study was funded by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream (LN) and the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network (LN). During the work presented in this article, JWG held the Scotiabank Chair in Child Health Research.

Table 3. Description of roles.

Role	Description	Illustrative Quotes
1. Caregiver	To care for the sibling with a neurodisability to carry out activities of daily living, such as daily hygiene and making food, while also making sure that the sibling continues with their routine schedule including going to school.	"I help him with his homework when I can. General hygiene, like brushing his teeth, taking him to the bathroom, showering, getting food, water, setting up his iPad so that he can play games." (Sister of a brother with cerebral palsy). "Sometimes I feel like his mini-parent, but not his parent. But I do, I've been protective of him since we were younger." (Older sister of a brother with autism spectrum disorder). "I've always been more of a caregiver... so in elementary school, we would go to school together. I would take the special transportation bus with her and I would bring her to class in the morning and then go to my class. And then after school, I would go get her and walk with her." (Older sister of a sister with a physical disability).
2. Role model/mentor	To complete activities related to school, such as tutoring, but to also provide mentorship in life such as making friends and new relationships.	"Yeah I do a lot of tutoring 'cause I tutor my cousins. I help my brother. And then I work at a tutoring centre." (Older sister of a brother with an intellectual disability). "Even with just like friends. He's like, like 'how do I kind of like figure that whole thing out? Like how do I like ask someone like 'oh like let's go hang out' like 'cause so I've kind of like, it shifted like as of like, I'm your big sister, I'm kind of more like a friend and like I've been through all of this so like I can help you with that kind of thing." (Older sister of a brother with autism spectrum disorder).
3. Protector	To provide a sense of security.	"All the things that were happening at school around them that I've noticed or like, you know, people teasing them, and you know, I tried my best to stick up for them and I would say something, but you know, because I was so small (laughter) and because I was so much younger, no one really at school took it seriously." (Younger sister of two older brothers with autism spectrum disorder).

4. Advocate	To provide information to explain to individuals, such as other family members, about disability. To begin to speak up about the importance of supports for their siblings with a neurodisability.	"To me, it's, it's quite spontaneous, quite natural. It's not something that I just plan to do. It, it just strikes up into conversation, so like it, it's quite natural. Or if my grandparents just like question about it, then, like if my grandparents just ask 'what's going on?' and stuff like that, then it's just natural to me. It's nothing like uh, there's nothing planned ahead." (Older brother of a brother with autism spectrum disorder). "Yeah, there is like, um, like a thing to sign up for, for talks with MPP with increasing public health funding. So um, so hopefully they'll do a related topic to my brother for this semester." (Older sister of a brother with autism spectrum disorder).
5. Supporter	To provide emotional support, such as being there for the sibling with a neurodisability when they are feeling overwhelmed or experiencing challenges. In some situations, this role would also involve supporting the sibling with a neurodisability in explaining information to parents and caregivers.	"Whenever she comes to me, it's more like emotional, so I would give her a hug or something. Let her know that I'm there." (Older brother of a sister with autism spectrum disorder). "I guess, he's, um, he gets discouraged very easily and sometimes overwhelmed, um, when he thinks that he can't succeed. So kind of always reminding him that, 'You know what, as long as you've done your best then um, like you've- you've given it your all so like you should be proud about that'". (Older sister of a brother with autism spectrum disorder). "I do things like, um, you know, [name of second oldest brother] and I, like I'll show him or I'll help him like look for something, even if it's just something simple like [name of second oldest brother] is saying, like, "Oh I want to tell Mom and Dad that" you know, I'm going to say this as an example like "I want to ask this for Christmas, can you- can you help me look for it". (Younger sister of a brother with autism spectrum disorder).
6. Friend	To have fun, have conversations and do activities together.	"We would really like to do things together and just have fun together and laugh. And he wants to have adventures. When we were little, we would build forts together and hang in the forts or like, just planning to do whatever outside with the neighbourhood kids." (Older sister of a brother with autism spectrum disorder). "At school, we'd be together, like at recesses or lunch time." (Younger sister of two brothers with cerebral palsy).

References

1. Stewart D. Transition to adult services for young people with disabilities: Current evidence to guide future research. Developmental Medicine and Child Neurology. 2009;51(Supp. 4):169–73.
2. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. Lancet Child Adolesc Health. 2018;2(3):223–8.
3. Freeman M, Stewart D, Cunningham CE, Gorter JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. Journal of Transition Medicine. 2018;1(1).
4. Blum RW. Improving transition for adolescents with special health care needs from pediatric to adult-centered health care: Introduction. Pediatrics. 2002;110(6 II):1301–3.
5. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. Journal of Applied Research in Intellectual Disabilities. 2018;31(3):423–34.
6. Atkin K, Tozer R. Personalisation, family relationships and autism: Conceptualising the role of adult siblings. Journal of Social Work. 2014;14(3):225–42.
7. Redquest BK, Tint A, Ries H, Goll E, Rossi B, Lunsky Y. Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2020;17(3):239–46.
8. Arnold CK, Heller T, Kramer J. Support needs of siblings of people with developmental disabilities. Intellectual and Developmental Disabilities. 2012;50(5):373–82.
9. Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, et al. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Frontiers in Rehabilitation Sciences. 2021 Oct 5;2:52.
10. Strategy for Patient-Oriented Research - Patient Engagement Framework - CIHR [Internet]. 2014 [cited 2018 Jan 14]. Available from: http://www.cihr-irsc.gc.ca/e/48413.html
11. Canadian Institutes of Health Research. Patient engagement [Internet]. 2019 [cited 2022 Apr 19]. Available from: https://cihr-irsc.gc.ca/e/45851.html
12. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: a scoping review of the ‘how’ and ‘what’ of patient engagement in health research. Health Research Policy and Systems. 2018;16(1):5.
13. Forsythe LP, Carman KL, Szydlowski V, Fayish L, Davidson L, Hickam DH, et al. Patient engagement in research: Early findings from the Patient-Centered Outcomes Research Institute. Health Affairs. 2019;38(3):359–67.
14. Canadian Institutes of Health Research. Strategy for Patient Oriented Research. Patient engagement framework [Internet]. 2014. Available from: http://www.cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf
15. Black A, Strain K, Wallsworth C, Charlton SG, Chang W, McNamee K, et al. What constitutes meaningful engagement for patients and families as partners on research teams? Journal of Health Services Research and Policy. 2018;23(3):158–67.
16. Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Protocol of the BEST SIBS study: A qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition. Journal of Transition Medicine. 2021;3(1):1–11.

17. CanChild Centre for Childhood Disability Research. BEST SIBS Study Poster. 2021.
18. CanChild Centre for Childhood Disability Research. BEST SIBS Study Recruitment Video. 2021.
19. CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study [Internet]. 2021 [cited 2022 Feb 4]. Available from: https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study
20. CHILD-BRIGHT Network. About Us: What is CHILD-BRIGHT? [Internet]. 2019 [cited 2022 Apr 18]. Available from: https//www.child-bright.ca/what-is-child-bright
21. Kids Brain Health Network. Overview [Internet]. 2021. [cited 2022 Apr 18]. Available from: https://kidsbrainhealth.ca/index.php/about/overview/
22. Gorter JW, Amaria K, Kovacs A, Rozenblum R, Thabane L, Galuppi B, et al. CHILD-BRIGHT READYorNot Brain-Based Disabilities Trial: Protocol of a randomised controlled trial (RCT) investigating the effectiveness of a patient-facing e-health intervention designed to enhance healthcare transition readiness in youth. BMJ Open. 2021;11:48756.
23. Patton MQ. Designing qualitative studies. In: Qualitative Evaluation and Research Methods. 2nd ed. Newbury Park: Sage Publications Inc.; 1990. p. 145–98.
24. Wang CC, Redwood-Jones YA. Photovoice ethics: perspectives from Flint Photovoice. Health Education & Behaviour. 2001;28(5):560–72.
25. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006;3(2):77–101.
26. Patton MQ. Enhancing the quality and credibility of qualitative analysis. Health Serv Res. 1999;34(5 Pt 2):1189–208.
27. Baxter P, Jack S. Qualitative case study methodology: Study design and implementation for novice researchers. The Qualitative Report. 2008;13(2):544–59.
28. Braun V, Clarke V. Reflecting on reflexive thematic analysis. https://doi.org/101080/2159676X20191628806. 2019;11(4):589–97.
29. Meleis AI, Sawyer LM, Im EO, Messias DK, Schumacher K. Experiencing transitions: an emerging middle-range theory. Advances in Nursing Science. 2000;23(1):12–28.
30. Bronfenbrenner U. Six theories of child development: Revised formulations and current issues. Vasta R, editor. London: Jessica Kingsley Publishers; 1992. 187–249 p.
31. Sankowski E. Responsibility of persons for their emotions. Canadian Journal of Philosophy. 1977;7(4):829–40.
32. McCormick M, Schleifer M. Responsibility for beliefs and emotions. Paideusis. 2020;15(1):75–85.
33. Burke MM, Taylor JL, Urbano R, Hodapp RM. Predictors of future caregiving by adult siblings of individuals with intellectual and developmental disabilities. American Journal on Intellectual and Developmental Disabilities. 2012;117(1):33–47.
34. Lee CE, Burke MM. Caregiving roles of siblings of adults with intellectual and developmental disabilities: a systematic review. Journal of Policy and Practice in Intellectual Disabilities. 2018;15(3):237–46.
35. Saxena M. Variables that can enhance and complicate sibling caregiving of individuals with intellectual and developmental disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2015;12(3):210–9.

36. Lee C, Burke MM, Stelter CR. Exploring the perspectives of parents and siblings toward future planning for individuals with intellectual and developmental disabilities. Intellectual and Developmental Disabilities. 2019;57(3):198–211.
37. Heller T, Arnold CK. Siblings of adults with developmental disabilities: Psychosocial outcomes, relationships, and future planning. Journal of Policy and Practice in Intellectual Disabilities. 2010;7(1):16–25.
38. Lee C eun, Burke MM, Arnold CK, Owen A. Compound sibling caregivers of individuals with intellectual and developmental disabilities. Journal of Applied Research in Intellectual Disabilities. 2020;33(5):1069–79.
39. Riley LD, Bowen C "Pokey." The sandwich generation: Challenges and coping strategies of multigenerational families. The Family Journal. 2005;13(1):52–8.
40. Griffiths DL, Unger DG. Views about planning for the future among parents and siblings of adults with mental retardation. Vol. 43. 1994.
41. Davys D, Mitchell D, Haigh C. Futures planning - adult sibling perspectives. British Journal of Learning Disabilities. 2015;43(3):219–26.
42. Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. Intellectual and Developmental Disabilities. 2009;47(3):208–19.
43. Bellefeuille S, Davis H, Havens J, Nguyen L. CanChild Luke's Legacy Family Research Rounds: The Art of Sibling Science [Internet]. 2021 [cited 2022 Mar 2]. Available from: https://vimeo.com/543787118
44. D'Arcy F, Flynn J, McCarthy Y, O'Connor C, Tierney E. Sibshops. An evaluation of an interagency model. Journal of Intellectual Disabilities. 2005;9(1):43–57.
45. Opening Hearts. SibTeens [Internet]. 2019 [cited 2021 Jun 2]. Available from: https://www.openinghearts.ca/sibteens/
46. The Sibling Collaborative [Internet]. [cited 2021 Jul 22]. Available from: https://sibcollab.ca/
47. Nguyen L, Bootsma J, Rezze B Di, Jack S, Ketelaar M, Gorter JW. Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: protocol of a scoping review. BMJ Open. 2021;11:53184.
48. Smith MJ, Greenberg JS, Seltzer MM. Siblings of adults with schizophrenia: Expectations about future caregiving roles. 2007;
49. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Canadian Journal of Community Mental Health. 2014;33(1):45–61.

CHAPTER SEVEN

TITLE: “Filling in the gap”: A qualitative case study about the influence of family relationships on identity formation of siblings of youth with a neurodisability

AUTHORS: Nguyen L, Jack SM, Davis H, Bellefeuille S, Arafeh D, Di Rezze B, Ketelaar M, & Gorter JW

JOURNAL: To be prepared for submission to a peer-reviewed journal focused on age-specific issues and life transitions of youth.

Abstract

Introduction: In families of children with a neurodisability, typically developing (TD) siblings have unique experiences that can shape their identity. There is limited information about the influence of sibling and family relationships on the identity formation of TD siblings of a sibling with a neurodisability. This study aims to understand the characteristics of relationships between TD siblings and their sibling with a neurodisability and parents/caregivers during childhood, adolescence, and emerging adulthood, and the influence of these relationships on identity formation of TD siblings.

Methods: As part of a patient-oriented research program, we engaged with the Sibling Youth Advisory Council in Canada. In this qualitative case study, data from 19 semi-structured interviews augmented by photo elicitation and graphic elicitation of relational maps was analyzed using reflexive thematic analysis.

Results: Nineteen sibling participants (mean age = 19.5 years, standard deviation = 5.0) reflected on their childhood, in which they were often playmates to their sibling with a neurodisability. During adolescence and emerging adulthood, they became closer with their sibling with a neurodisability, provided support or sought advice from their TD siblings, and increased communication with their parents about how to interact and care for their sibling with a neurodisability. These relationships influenced the identity of sibling participants, in which they considered how to balance their life choices with family needs.

Conclusions: Siblings form a unique identity and require support in this process. The support of all family members can create a positive family environment that can lead better health outcomes for adolescents with a neurodisability.

Keywords: youth, sibling, neurodisability, identity formation, qualitative

Introduction

Siblings are important members of the family, and the nature of how sibling relationships look like can evolve and change over time (1). Siblings typically grow up together with a fundamental presence in each other lives that can look different at each developmental stage. During childhood, siblings can experience both positive and negative emotions with roles such as a playmate or supporter (1,2). They may also have conflicts such as disputes over toys or needs for parental attention (3,4). During adolescence, youth begin to develop their own identity by exploring interests that may result in spending less time in activities with their siblings (5). However, some youth may refer to their older siblings as sources of support regarding personal and family issues (6). When youth enter into emerging adulthood between 18 to 25 years old, the interactions with their siblings is often voluntary (7). Young adults may feel closer with their siblings and continue to seek support from them (8,9).

The formation of identity is a universal experience that begins during adolescence and continues into emerging adulthood (10–13). Identity formation is defined as the process of developing a distinct personality, in which the individual is known for specific characteristics (10). The process of forming an identity includes an active stage of exploration, which refers to the questioning and weighing of different options of identity (14). Adolescents may experience a dynamic balance between commitment levels and uncertainty with their identity (12). They may form commitments, but also continue to choose or reconsider their commitments through a deeper exploration of their identity (14–16).

Furthermore, adolescents are beginning to understand how their identity is formed by situations in their environment (17). The interaction between the person and the environment that can affect the development of a person (18). In families of one or more children with a disability, typically developing (TD) siblings may have different experiences of their sibling relationship that can influence the formation of their identity. During adolescence, they may begin to develop skills in perspective taking in which they recognize how to communicate and connect with their sibling with a neurodisability (19,20). They may also start to recognize the needs of the family, for example, cancelling their own plans to stay at home to care for their sibling with a neurodisability when their parents were not available (21). As TD siblings enter emerging adulthood, they experience situations and conflicts that their peers are also facing. For example, they may leave home to live independently while maintaining close relationships with their family. However, TD siblings face additional tension in their identity formation because they may consider how to balance their identity and roles at home. The roles that adult TD siblings have described include being a caregiver, advocate, or informal service coordinator (22,23).

A previous review and qualitative analysis of online resources to support siblings of individuals with a chronic health condition identified that siblings often discussed about their identity and roles in blogs and interviews (24). Siblings shared in blogs and interviews that their experiences of growing up with their sibling with a neurodisability motivated them to pursue careers, such as healthcare professions or research, to support children with a chronic health condition. In our conversations with young adult siblings with lived experiences, they shared how their unique identity was continuously influenced by their relationship with their sibling with a disability. For example, while their sibling with a neurodisability was transitioning to adulthood, they were also experiencing their own transition to adulthood. They developed skills, such as empathy and increased responsibility to be present for their family, and they were also learning how to develop their identity outside of the family such as in their education and careers. Although TD siblings are influenced by their relationship and roles with their sibling with a disability, there is limited information about how these experiences affects their identity formation (25).

The purpose of this study is to describe the characteristics of relationships between TD siblings and siblings with a neurodisability and other members in their immediate family during childhood, adolescence, and emerging adulthood. Then, TD siblings' perceptions about the various ways in which these relationships influence the development of their personal identity are also described.

Methods

Engagement of siblings with lived experiences

The representation of lived experiences of siblings in research can help to increase the relevance and applicability of the study to them (26–28). In this study, we established and partnered with the Sibling Youth Advisory Council (SibYAC) comprised of young adult siblings (ages 21-28 years old) with a sibling with a disability in Canada. Our qualitative case study titled, BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study, aimed to understand the roles and responsibilities of siblings of youth with a neurodisability (29). This study emerged based on the lived experiences of the SibYAC who described how they were continuously forming their identity that is influenced by development of their relationships with the sibling with a neurodisability and sibling.

Case study

This qualitative descriptive study aimed to describe the experiences of TD siblings in how their relationships with their sibling with a neurodisability and family influenced their identity formation. The experiences of TD siblings will be analyzed as a 'single case' bounded by geographical location and time to ensure that the analysis is feasible within scope. This study focused on the geographical location of Ontario to ensure that all TD siblings had similar experiences in access to services, such as healthcare. This study expanded on the BEST SIBS Study to focus on the experiences of TD siblings across the developmental transition time periods of childhood, adolescence, and emerging adulthood. Details of the BEST SIBS Study is described in our published protocol (29).

Participant recruitment

Participants met the following inclusion criteria: 1) were between 14 to 40 years old; 2) has a sibling between 14 to 21 years old with a diagnosis of a childhood-onset neurodisability; and 3) speaks English. The study recruitment materials included a study website (18), as well as a study poster (19) and video that was co-created with the SibYAC (20). Study recruitment materials were shared on social media platforms, through networks from the research team and disability-specific organizations CHILD-BRIGHT Network (30) and Kids Brain Health Network (31). This study also recruited potential siblings of the brother or sister with a neurodisability from Ontario who participated in a multi-regional Canadian randomized controlled trial (21). Snowball sampling was used, in which participants could share the study recruitment materials with other siblings who may be interested in participating (22). Data collection and data analysis occurred in parallel, and participants were recruited until a detailed description of key themes was obtained. Participants were recruited from February 2020 to July 2021. This study will refer to ‘sibling participant’ instead of ‘TD sibling’ when describing the results, as a sibling was not excluded if they had a neurodisability.

Ethical approval

Ethics approval was obtained from the Hamilton Integrated Research Ethics Board (Project #7932). A three-step process was used to obtain consent (32). First, all participants were asked to provide consent to participate in the interviews. Second, participants were asked to share photographs which could include their parents and/or sibling. Third, participants were asked for permission to have the photographs shared on specific platforms such as for study publication, workshops, or conferences. The second and third steps were optional. To provide a welcoming and safe environment for participants, the interviewer (LN) asked if there was anything that could be considered during the interview to make them feel more comfortable. Participants could also choose not to answer any questions or stop the interview at any time.

Data collection

Data was collected with semi-structured, 1:1 in-depth interviews augmented with photo elicitation and graphic elicitation of relational maps (25). Due to public health measures and physical distancing guidelines to reduce the transmission of SARS-Cov-2, the first author (LN) conducted all interviews via a web conference platform. During the interviews, photo elicitation techniques were used to deepen the conversation with sibling participants. They were asked to share 3-5 pre-existing photographs that described their sibling experiences, either by bringing the photographs to the interview or uploading the photographs onto a secure cloud storage. At the beginning of the interviews, participants were asked to draw a family tree diagram of members in their intermediate family, a technique known as graphic elicitation of relational maps (25). Participants could verbally describe or show their completed family tree diagram using their web camera. All participants were asked an initial broad question of ‘tell me about your sibling relationship'. Participants were then asked to select the order that they would like to share the photographs and to describe the photographs based on prompts such as who is in the photographs and what do the photographs mean to them (33,34).

Data analysis

The verbal descriptions of the photographs and family tree diagrams were analyzed as part of the interview transcripts. Reflexive thematic analysis was used to analyze the interviews with siblings (35).

A conceptual framework was applied to ensure that there were boundaries about the focus of analysis (36). This study focused on the analysis of a subset of data, specifically the sibling participants' development of relationships with their sibling with a neurodisability and family, and its influence on their identity formation. A conceptual framework was developed to guide data analysis using two theories:

1. *Transition theory* (37)*:* This theory describes the transition experience, including types of transitions, as well as the properties, facilitators and inhibitors, and outcomes of transitions. Data analysis focused on the transition conditions, primarily on the personal conditions such as the meaning of transition that sibling participants experienced.
2. *Bioecological systems theory* (38): This theory describes person-environment transactions. The application of this theory helped to focus the analysis on the mesosystem, which included the relationships between the sibling participant and their intermediate family including their sibling with a neurodisability, other siblings (if applicable), and parents and/or caregivers. These relationships can influence how sibling participants form their personal identity.

The propositions included in this conceptual framework provided guidance on the factors to analyze that could influence the sibling relationship. These propositions included gender, age, and health of the sibling participant and sibling with a neurodisability. In addition to these propositions, identity formation of the TD siblings will be further analyzed based on three components: distinctiveness, in which an individual sees their self as unique from others; coherence, with a perception of the self as similar across different domains including education, work, and relationships; and continuity, with a perception of being the same self over time (39).

Field notes written by LN after each interview were reviewed to inform the development of codes, categories, and preliminary themes. Meetings were held with stakeholders, comprised of the SibYAC and five graduate students, to review a summary of preliminary themes as a form of analyst triangulation and peer debriefing to enhance data credibility (40). Stakeholders were from multidisciplinary backgrounds, including cognitive psychology, education, nursing, occupational therapy, physiatry, rehabilitation, user experience design, patient-oriented research, and lived experiences.

Results

There were nineteen sibling participants (mean age = 19.5 years, standard deviation [SD] = 5.0) with 12 females and 7 males. Three participants self-declared that they had a neurodisability. There were fourteen participants who described their relationship to one sibling with a neurodisability and five participants who described their relationship to two siblings with a neurodisability. There was a total of twenty unique siblings with a neurodisability (mean age = 16.9 years, SD = 2.1) with 5 females and 15 males described during the interviews. The type of neurodisability of the sibling included autism spectrum disorder (n = 12), cerebral palsy (n = 4), hereditary spastic paraplegia (n = 1), intellectual disorder (n = 1), attention deficit hyperactivity disorder (n = 1), and epilepsy (n = 1). The interviews with sibling participants ranged from 45 to 104 minutes. Table 1 describes the characteristics of the siblings in the family.

Table 1. Characteristics of siblings in the family.

Characteristics	**Total number**
Sibling dyad (sibling participant-sibling with a neurodisability)	
Sister-brother	11
Sister-sister	3
Brother-sister	4
Brother-brother	6
Birth order (sibling participants to their sibling with a neurodisability)	
Older sister, brother	7
Older sister, sister	3
Older brother, sister	4
Older brother, brother	2
Younger sister, brother	4
Younger sister, sister	0
Younger brother, sister	0
Younger brother, brother	4
Number of siblings with a neurodisability in the family	
1 sibling with a neurodisability	15
2+ siblings with a neurodisability	4
Total number of siblings in the family	
2 siblings	8
3 siblings	6
4+ siblings	5

Sibling participants spoke about their relationships with their siblings within the context of the broader family structure. In their narratives, as well as within the relational maps, sibling participants discussed about the different relationships that exist within their immediate family including with their sibling(s) with a neurodisability, other TD siblings, and parents and/or caregivers. Sibling participants further shared how these relationships evolved during their developmental stages of childhood, adolescence, and emerging adulthood. Being in a relationship with a sibling with a neurodisability also profoundly influenced the sibling participants' perceptions of how they developed and formed their personal identity. Siblings described how a part of their identity was ‘filling in the gap’ similar to a puzzle piece in the family by taking on different roles where that they were most needed to support the whole family through the developmental transitions of themselves and their family (See Figure 1).

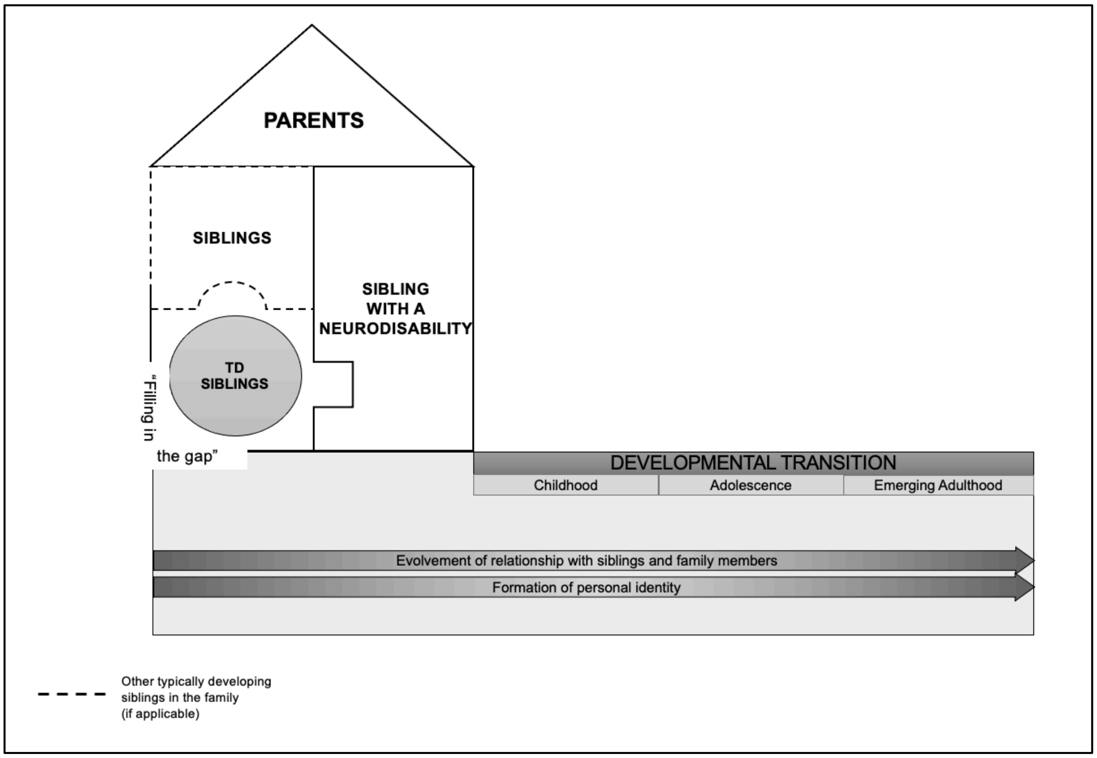

Figure 1. *The development of the relationships that sibling participants with their sibling with a neurodisability and family members, which influences the formation of personal identity.*

Childhood

Family Relationships: Exploring and recognizing the uniqueness of sibling and family relationships

As sibling participants reflected on their childhood, they described how they were playmates with their sibling with a neurodisability. Some siblings described how they would fight over small conflicts such as toys or making fun of each other. During interactions with their sibling with a neurodisability, sibling participants always knew that there was something different, but they did not fully understand the neurodisability of their sibling until their parents provided an explanation. Sibling participants often received the explanation about a neurodisability from their parents when their sibling was first diagnosed. While they continued to ask their parents question about what a neurodisability means, they all stated that they loved their sibling with a neurodisability for who they were.

Identity Formation: Relating individual identity to the sibling and family

During childhood, a distinct sibling identity was not clearly formulated yet. Sibling participants were all considering how their individual selves were related to their sibling with a neurodisability. A sibling participant described how her sibling relationship was a part of her normal life: “I know that our family is not exactly what you would call like a normal family, but to me, it’s my normal and you know, I wouldn’t, I wouldn’t trade it for anything really” (Older sister of a brother with cerebral palsy). Sibling participants recognized that the relationship that they have with their sibling with a neurodisability was unique and distinct from other TD sibling relationships.

Adolescence

Family Relationships: Understanding and navigating different family perspectives

During adolescence, sibling participants described how the nature of their relationships with their sibling with a neurodisability, other TD siblings, and parents began to change. Both the sibling participants and siblings with a neurodisability began to discuss topics that were important to them. For example, the siblings with a neurodisability might turn to their siblings for advice about personal relationships with friends and significant partners or future career choices after graduating from high school. Not only did the siblings participants offer advice, but the siblings with a neurodisability might also seek ways to offer care and support in their own way. Some siblings with a neurodisability might offer a different perspective to a problem that the sibling participants might have:

> A big thing that I found is that he looks at things in such a different way that I do. So I am very analytical... I like it when things are like black and white... so when things are grey, I'm like 'This is, I'm like there's so many options, like I don't like it'. So he just approaches things from a completely different lens.' (Older sister of a brother with autism spectrum disorder).

Sibling participants began to connect more with their other TD siblings. For example, adolescent sibling participants turned to their older siblings for support, such as how to navigate the challenges around the behaviour of a sibling with a neurodisability. For example, during adolescence, the sibling with a neurodisability might also need their space and independence from the family. The behaviour of the sibling with a neurodisability could sometimes be hurtful as they might distance themselves from the family through aggression or verbal comments. Older siblings, including those who have a neurodisability, might have had similar behaviours or experiences and so, they could provide advice to their younger siblings.

Sibling participants continued to have conversations with their parents about the behaviour of their sibling with a neurodisability. An older sister described the questions that she asked her mother when she noticed the challenging behaviours that her younger brother with a neurodisability had:

> Lately, he's been getting really angry about things... so if that ever happens, we'll talk about it after and my mom will just talk about what happened... my mom's like 'Oh, like maybe next time, don't do that because that made him get more angry' (Older sister of a brother with an intellectual disability).

Sibling participants were also beginning to communicate with their parents about their own needs while also advocating for the needs of their sibling with a neurodisability. Sibling participants described that their parents needed to recognize that each sibling was different with individual strengths, needs, and areas of improvement.

Identity Formation: Becoming independent and self-reliant

Based on ongoing conversations that sibling participants had with their sibling with a neurodisability and parents affected their personal identity, they recognize who they needed to be in their sibling identity when they are with their family. There was coherence in their sibling identity, in which sibling participants recognized when they needed to take a step back to ensure that the needs of other family members were met first. Sometimes, stepping back meant that they made time to have conversations with their other siblings, such as providing advice and support. Sibling participants also described inherent compromises that they had to make, which indirectly affected the identity of sibling participants. For example, some sibling participants described that they had to walk to the community centre or take the bus by themselves to attend the activities or stop taking part in the activities because their parents could not pick them up. They also described how they would have to make plans to socialize with friends in advance to ensure that there was a family member to take care of their sibling with a neurodisability. Across all their domains in life, with school, friends, and family, sibling participants often placed the needs of their sibling with a neurodisability and family as the priority.

They also identified their identity as a student as a priority, as many sibling participants were focused on completing their schoolwork and exploring their interests through extracurricular activities. While they were developing their identity as a student, they continued to consider how they could balance their studies with their responsibilities as a sibling to their sibling with a neurodisability. While some of these responsibilities were similar to their peers, such as walking to school and babysitting them after school, other types of care tasks related to the sibling with a neurodisability required more time and attention. Sibling participants had to explain to their sibling with a neurodisability when they were not able to assist with homework because they needed to focus on their own studies. For example, sibling participants shared about wanting to help their sibling with a neurodisability with their schoolwork but they needed to also focus on their own studies. An older sister in university described how she needed to communicate with her brother with a neurodisability when she was not able to help him with his homework:

> I usually have to kind of emphasize how I'm also in school and I also need to uh, like, sometimes I'm cooped all day in group calls and everything and I might not be able to help him at that moment, but kind of reassuring him that once I have time, 'Don't worry, I'll help you.' And then kind of reassuring him that he's more than capable of finishing the assignments. (Older sister of a brother with autism spectrum disorder).

Adolescent sibling participants were learning how to explore their personal interests and focus on their studies, while considering the needs of their sibling with a neurodisability.

Emerging adulthood

Development of family relationships: Strengthening connections with family

All sibling participants described a stage of having entered or preparing to enter emerging adulthood when they had less time to spend with their sibling with a neurodisability. Sibling participants might have other commitments, such as moving away from home to attend university or working part-time jobs. Communication often looked different where instead of conversations in-person, there were more texts, pictures, or videos sent between siblings. Older female siblings often sent daily messages or pictures to stay connected while they were physically living away from their sibling with a neurodisability. Some siblings preferred to connect in person, which allowed them to strengthen their bond with each other when they met.

All sibling participants described how the relationship with their sibling with a neurodisability affected their choices, such as with post-secondary education and careers. They often made plans that centred around the care of their family as the priority. A sibling participant her worry for her mother when she moved away for university: “I hope that it’s not just all my mom because before it was just all my mom’s [for example, work in finding resources] so I just hope that people will help her” (Older sister of a brother with an intellectual disability). If there were other siblings in the family, sibling participants expressed how they had ongoing discussions about changes in role when they moved away. These roles included who would provide continue to provide care for their sibling with a neurodisability, such as taking them to healthcare appointments or reminding them to take medications.

The active role that sibling participants wanted to have with their sibling with a neurodisability encouraged them to share their perspectives on certain decisions that parents made for the sibling with a neurodisability. Some decisions included the types of treatments, therapies, or activities that the sibling with a neurodisability engaged in. When there were disagreements, sibling participants recognized that they could share their perspectives, but they also needed to take a step back as the legal decisions were up to their parents.

Identity Formation: Reconciling different identities

Sibling participants all described the love that they had with their sibling with a neurodisability, in which they wanted to take on certain roles to alleviate the burden in the family. When they placed their family needs as a priority, the continuity of their sibling identity to be independent continued during emerging adulthood. While some sibling participants might want to go to their parents for support about their personal problems, they also did not want to be an additional burden. For the eldest sibling participants, they especially recognized that their parents might have to care for their younger siblings and siblings with a neurodisability. An older brother of a sister with a neurodisability reflects on how he grew to become independent:

> Sometimes I think I've probably had moments when I've had my own issues that I want to bring home and tell my parents about. But in a home environment, I can see my parents were already taxed and stressed, dealing with [sibling with a neurodisability] and her struggles that I was just like, 'Okay, I'm not going to bring it up'. Or I'm just going to try to problem solve it myself. (Older brother of a sister with attention deficit hyperactivity disorder).

Sibling participants understood that there were priorities in the family, and the eldest siblings often became independent by learning how to problem-solve on their own.

During emerging adulthood, sibling participants were also developing their professional identity and career. Many sibling participants described how they were influenced to enter professional careers that were influenced by their relationship with their sibling with a neurodisability. For example, some sibling participants chose healthcare professions that could support individuals with a disability. Other sibling participants identified that they were motivated to enter professional careers that could offer financial stability, which would then allow them to support their sibling with a neurodisability in the future.

In their social identity, sibling participants described the relationships that they had with their peers and romantic partners. Sibling participants recognized that they were distinct from their peers, and they were most able to bond with peers who had similar experiences of having a sibling with a neurodisability. Some sibling participants joined online groups to connect with other siblings of individuals with a neurodisability, which offered a place to share their concerns and worries about their sibling with a neurodisability. However, they also needed to find a balance in which they did not want to feel overwhelmed by the discussions in an online forum. The relationship with the sibling with a neurodisability continued to influence the romantic relationships that sibling participants had. Many sibling participants identified that it was critical that their romantic partner was able to get along with their sibling with a neurodisability. They often explained to their partners about the neurodisability before introducing their partners to their sibling with a neurodisability.

Among all sibling participants, there was coherence in how their identity as a sibling will be at the forefront across different aspects of their life including their professional and social life. For some sibling participants, a part of this coherence of their sibling identity is prioritizing the needs of the sibling with a neurodisability and family. All sibling participants identified that their sibling identity would continue across time, and that their sibling relationship will be an important part of their life.

Discussion

This study focused on understanding how the development of relationships that TD siblings had with their sibling with a neurodisability, other TD siblings, and parents influenced the formation of their personal identity. Sibling participants described how they were ‘filling in the gap’ in the family with responsibilities that they could take on based on their stage of transition in life. They described how they cared for their sibling with a neurodisability, provided support or asked for advice from their other TD siblings, and referred to their parents for information about a neurodisability or as the final decision-maker in the family. Throughout adolescence and emerging adulthood, siblings were learning how to reconcile their different identities including as a sibling and student, as well as socially and professionally.

Sibling participants described how growing up with their sibling with a neurodisability was a part of their normal life. Similar experiences have been described in the literature, in which

TD siblings are growing up with their sibling with a neurodisability and they will share life events together (41,42). The changes in the level of closeness between siblings over time can be described as a U-shaped curve (19). Siblings often experience the highest and closest relationship during childhood. Similar to how sibling participants reflected on their childhood experiences, TD siblings have described how they understand the preferences and feelings of their sibling with a neurodisability, but also have difficulties with adapting to challenging behaviours (41). During adolescence and emerging adulthood, there may be a decrease in closeness between siblings with and without a neurodisability (19). TD siblings have their personal identity and experience transitions themselves, such as moving away from home, pursuing postsecondary education, or having a job (43).

This study provides a further contribution to the literature to understand in-depth about how TD siblings process their family relationships and form an identity. Based on the narratives shared from sibling participants, their identity formation has similarities and differences from their TD peers. During adolescence, sibling participants have similar experiences to their TD peers as they begin to explore their interests, develop perspective-taking skills, and become independent (20). However, sibling participants have additional experiences of growing up with their sibling with a disability that further influences their identity formation. Across all sibling participants, their experiences and family relationships influences the three components of identity (39): distinctiveness as their identity as a sibling is different and unique from their peers; coherence, as they identify that being a sibling is important across all aspects of their life; continuity, in which they will continue to assume their sibling role and care for their sibling with a neurodisability throughout their life.

An important component to the identity formation was how sibling participants made sense of their role as a sibling. During childhood and adolescence, sibling participants needed to understand the neurodisability and learn how to interact with their sibling with a neurodisability. Similar to the experiences from sibling participants, an integrative review synthesized studies about sibling adjustment to childhood chronic illness that identified how TD siblings learned knowledge about the illness from their parents (44). While TD siblings expressed wanting to learn more information about the illness of their sibling, parents decided how much information to share based on the age of the TD siblings and the type of diagnosis (44–46). Sibling participants further described how their mothers often provided information and resolved conflicts in the sibling relationship. In the literature, mothers tend to have primary responsibility to care for their children with a disability while also advocating for their children with professionals and service providers (47,48). This study further highlights the significant role that mothers have for the well-being of the whole family, including offering support to TD siblings.

Sibling participants shared how a part of their identity was 'filling in the gap' in the family and to be independent. Sibling participants learned how to problem-solve their personal problems to alleviate burden in the family. They also expressed a need to fulfill roles based on the family needs, such as providing assistance and care to their sibling with a neurodisability. This need to fulfill the sibling role based on the family needs can be similar may be associated with having a high internal locus of control, which is the degree to which an individual has control over life events (49). Emerging adult siblings of individuals with a disability have been identified to have a higher locus of control (49). The SibYAC has shared about the need to have control over life events in their family. For example, the SibYAC shared their role as a sibling to make plans and ensure that family outings were accessible for their sibling with a disability.

Strengths and limitations

A strength of this study is the involvement with the SibYAC as partners, and their lived experiences and expertise contributed to the analysis of the study findings. Another strength is the use of photo elicitation and graphic elicitation of relational maps, which encouraged sibling participants to reflect on their experiences and share rich details of their stories throughout childhood, adolescence, and emerging adulthood during the interviews. A study limitation is that the siblings who participated in this study may also have an increased role to their sibling with a neurodisability and were interested in reflecting about their experiences with a neurodisability compared to siblings who did not participate in this study. However, the strategies used for data analysis allowed for an in-depth understanding about the perceptions of sibling participants on their relationship with their sibling with a neurodisability, the influence of this relationship on their identity formation. An additional limitation is that all sibling participants grew up in the same household, and most sibling participants were older than the sibling with a neurodisability who were raised by their biological parents. Siblings who grew up in foster care, institutional or out-of-home care, or with their step- or adoptive family may have different experiences (50–53). While these demographic characteristics of the family may impact the nature and dynamic of the sibling relationships, the experiences shared by sibling participants were analyzed in-depth.

Implications for healthcare and future directions for research

There were discussions with the SibYAC and research team about the implications of the findings for healthcare and future directions for research. This study highlighted that siblings also require support for the positive formation of their identity. Siblings have an important role in the family, and healthcare professionals could consider the family context with a life course approach when providing care to the youth with a neurodisability. The person-environment interactions along with the influence of relationships can lead to outcomes for youth with a neurodisability (18). The ecological systems theory also illustrates the interactions that children and youth have in multiple systems, including the family ecosystem (38). A positive family environment, where all family members are supported in their health and well-being, could lead to better health outcomes for children and youth with a neurodisability (18).

Further research should be conducted to better understand the relationship of siblings from a variety of family dynamics, such as different family types with half siblings and step siblings, and siblings who grow up in separate households. Other demographics of families, such as socioeconomic status, can also influence the sibling relationship (54,55). Healthcare services could be tailored to the individual needs and circumstances of each family (56). Families of youth with a disability have emphasized the importance for healthcare professionals to understand the complexities of family circumstances that could affect their involvement in healthcare (56). Considerations can be incorporated into the health system to better support the child and family, as families continue to navigate the needs of their child with a neurodisability.

Additional research can be conducted about the influence of culture on sibling relationships. The cultural context can affect how TD siblings experience their relationship with their sibling with a neurodisability (57,58). Some cultures emphasize the importance of family, in which family members can depend on each other (59,60). Children of immigrant families, for example, may be expected to take on more caregiving roles due to a lack of financial resources or social support (61,62).

Conclusion

TD siblings are growing up alongside their sibling with a neurodisability. While the sibling relationship evolves over time based on shared experiences, TD siblings are also developing their personal identity. Communication served as the foundation that influenced the ongoing development of family relationships that TD siblings had, including with their sibling with a neurodisability, other siblings, and parents/caregivers. Resources and services could be offered to support the whole family.

Acknowledgements

We are grateful to the siblings who participate in this study. We gratefully acknowledge our partnership with the Sibling Youth Advisory Council throughout all study phases, and some members chose to be involved in a consultation role instead of as co-authors. We also thank our colleagues who were involved with the peer debriefing process: Carrie Bullard, Shelby Deibert, Tessa Elliott, Paige Gehrke, and Carly Whitmore.

Disclosure statement

The authors have no conflicts of interest.

Funding

This study was funded by the Canadian Institutes of Health Research Patient-Oriented Research Award – Transition to Leadership Stream (LN) and the Graduate Student Fellowship in Patient-Oriented Research through the CHILD-BRIGHT Network (LN). During the work presented in this article, JWG held the Scotiabank Chair in Child Health Research.

References

1. Scharf M, Shulman S, Avigad-Spitz L. Sibling relationships in emerging adulthood and in adolescence. Journal of Adolescent Research. 2005;20(1):64–90.
2. Hughes C, McHarg G, White N. Sibling influences on prosocial behavior. Current Opinion in Psychology. 2018;20:96–101.
3. Ross HS. Negotiating principles of entitlement in sibling property disputes. Developmental Psychology. 1996;32(1):90–101.
4. Lashewicz B, Keating N. Tensions among siblings in parent care. European Journal of Ageing. 2009;6(2):127–35.
5. Dunn J, Slomkowski C, Beardsall L. Sibling relationships from the preschool period through middle childhood and early adolescence. Developmental Psychology. 1994;30(3):315–24.
6. Tucker CJ, McHale SM, Crouter AC. Conditions of sibling support in adolescence. Journal of Family Psychology. 2001;15(2):254–71.
7. Stewart RB, Kozak AL, Tingley LM, Goddard JM, Blake EM, Cassel WA. Adult sibling relationships: Validation of a typology. Pers Relatsh. 2001;8(3):299–324.
8. Teti DM. Retrospect and prospect in the psychological study of sibling relationships. McHale JP, Grolnick WS, editors. Mahwah, NJ: Lawrence Erlbaum; 2002. 193–224 p.
9. Cicirelli VG. A comparison of college women's feelings toward their siblings and parents. Journal of Marriage and the Family. 1980;42(1):111.
10. Sharma S. Self, identity and culture. In: Interdisciplinary Perspectives on Consciousness and the Self. New Delhi: Springer India; 2014. p. 117–24.
11. Erikson E. Childhood and society. New York: Norton; 1963.
12. Erikson E. Identity: Youth and crisis. New York: Norton; 1968.
13. Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. American Psychologist. 2000;55(5):469–80.
14. Marcia JE. Development and validation of ego identity status. Journal of Personality and Social Psychology. 1966;3(5):551–8.
15. Crocetti E, Rubini M, Meeus W. Capturing the dynamics of identity formation in various ethnic groups: Development and validation of a three-dimensional model. Journal of Adolescence. 2008;31:207–22.
16. Meeus W. Studies on identity development in adolescence: An overview of research and some new data. Journal of Youth and Adolescence1. 1996;25(5):569–98.
17. Cinoğlu H, Arıkan Y. Self, identity and identity formation: From the perspectives of three major theories. Journal of Human Sciences. 2002;9(2):1114–31.
18. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Canadian Journal of Community Mental Health. 2014;33(1):45–61.
19. Neyer FJ. Twin relationships in old age: A developmental perspective. Journal of Social and Personal Relationships. 2002;19:55–177.
20. Ontario Ministry of Children, Community and Social Services. Stepping stones: A resource on youth development [Internet]. 2012 [cited 2022 Apr 22]. Available from: https://children.gov.on.ca/htdocs/English/documents/youthopportunities/steppingstones/SteppingStones.pdf

21. Corsano P, Musetti A, Guidotti L, Capelli F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. Journal of Intellectual and Developmental Disability. 2017;42(2):151–61.
22. Bigby C. Parental substitutes? The role of siblings in the lives of older people with intellectual disability. Journal of Gerontological Social Work. 1997 Jun;29(1):3–21.
23. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. Journal of Applied Research in Intellectual Disabilities. 2018;31(3):423–34.
24. Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, et al. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Frontiers in Rehabilitation Sciences. 2021 Oct 5;2:52.
25. McHale SM, Updegraff KA, Feinberg ME. Siblings of youth with autism spectrum disorders: Theoretical perspectives on sibling relationships and individual adjustment. Journal of Autism and Developmental Disorders. 2016;46(2):589–602.
26. Canadian Institutes of Health Research. Patient engagement [Internet]. 2019 [cited 2022 Apr 19]. Available from: https://cihr-irsc.gc.ca/e/45851.html
27. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: a scoping review of the 'how' and 'what' of patient engagement in health research. Health Research Policy and Systems. 2018;16(1):5.
28. Forsythe LP, Carman KL, Szydlowski V, Fayish L, Davidson L, Hickam DH, et al. Patient engagement in research: Early findings from the Patient-Centered Outcomes Research Institute. Health Affairs. 2019;38(3):359–67.
29. Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Protocol of the BEST SIBS study: A qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition. Journal of Transition Medicine. 2021;3(1):1–11.
30. CHILD-BRIGHT Network. About Us: What is CHILD-BRIGHT? [Internet]. 2019 [cited 2022 Apr 18]. Available from: https://www.child-bright.ca/what-is-child-bright
31. Kids Brain Health Network. Overview [Internet]. 2021 [cited 2022 Apr 18]. Available from: https://kidsbrainhealth.ca/index.php/about/overview/
32. Wang CC, Redwood-Jones YA. Photovoice ethics: perspectives from Flint Photovoice. Health Education & Behaviour. 2001;28(5):560–72.
33. Wang, Burris. Photovoice: concept, methodology, and use for participatory needs assessment. Health Educ Behav. 1997;24(3):369–87.
34. Wang CC. Photovoice: A participatory action research strategy applied to women's health. Journal of Women's Health. 1999;8(2):185–92.
35. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006;3(2):77–101.
36. Baxter P, Jack S. Qualitative case study methodology: Study design and implementation for novice researchers. The Qualitative Report. 2008;13(2):544–59.
37. Meleis AI, Sawyer LM, Im EO, Messias DK, Schumacher K. Experiencing transitions: an emerging middle-range theory. Advances in Nursing Science. 2000;23(1):12–28.
38. Bronfenbrenner U. Six theories of child development: Revised formulations and current issues. Vasta R, editor. London: Jessica Kingsley Publishers; 1992. 187–249 p.

39. van Doeselaar L, Becht AI, Klimstra TA, Meeus WHJ. A review and integration of three key components of identity development. European Psychologist. 2018 Oct 7;23(4):278–88.
40. Patton MQ. Enhancing the quality and credibility of qualitative analysis. Health Serv Res. 1999;34(5 Pt 2):1189–208.
41. Luijkx J, van der Putten AAJ, Vlaskamp C. "I love my sister, but sometimes I don't": A qualitative study into the experiences of siblings of a child with profound intellectual and multiple disabilities†. Journal of Intellectual and Developmental Disability. 2016;41(4):279–88.
42. Avieli H, Band-Winterstein T, Araten Bergman T. Sibling relationships over the life course: growing up with a disability. Qualitative Health Research. 2019;29(12):1739–50.
43. Hodapp RM, Sanderson KA, Meskis SA, Casale EG. Adult siblings of persons with intellectual disabilities: Past, present, and future. International Review of Research in Developmental Disabilities. 2017;53:163–202.
44. Lummer-Aikey S, Goldstein S. Sibling adjustment to childhood chronic illness: An integrative review. Journal of Family Nursing. 2021;27(2):136–53.
45. Plumridge G, Metcalfe A, Coad J, Gill P. Parents' communication with siblings of children affected by an inherited genetic condition. Journal of Genetic Counseling. 2011;20(4):374–83.
46. Williams PD, Williams AR, Graff JC, Hanson S, Stanton A, Hafeman C, et al. Interrelationships among variables affecting well siblings and mothers in families of children with a chronic illness or disability. Journal of Behavioral Medicine. 2002;25(5):411–24.
47. Ryan S, Runswick-Cole K. Repositioning mothers: mothers, disabled children and disability studies. Disability & Society. 2008;23(3):199–210.
48. Traustadottir R. Mothers who care: Gender, disability, and family life. Journal of Family Issues. 1991;12(2):211–28.
49. Burton SL, Parks AL. Self-esteem, locus of control, and career aspirations of college-age siblings of individuals with disabilities. Social Work Research. 1994;18(3):178–85.
50. Hunsley JL, Ekas N V., Crawley RD. An exploratory study of the impact of adoption on adoptive siblings. Journal of Child and Family Studies. 2021;30(1):311–24.
51. Reed JA. We live here too: Birth children's perspectives on fostering someone with learning disabilities. Children & Society. 2007;8(2):164–73.
52. Thompson H, McPherson S. The experience of living with a foster sibling, as described by the birth children of foster carers: a thematic analysis of the literature. Adoption & Fostering. 2011;35(2):49–61.
53. Mirfin-veitch B, Bray Nicola Ross A. "'It was the hardest and most painful decision of my life!'": Seeking permanent out-of-home placement for sons and daughters with intellectual disabilities* Impetus for the deinstitutionalisation. Journal of Intellectual & Developmental Disability. 1985;28(2):99–111.
54. Mulroy S, Robertson L, Aiberti K, Leonard H, Bower C. The impact of having a sibling with an intellectual disability: parental perspectives in two disorders. Journal of Intellectual Disability Research. 2008;52(3):216–29.
55. Giallo R, Gavidia-Payne S. Child, parent and family factors as predictors of adjustment for siblings of children with a disability. Journal of Intellectual Disability Research. 2006 Dec;50(12):937–48.

56. Allen D, Scarinci N, Hickson L. The nature of patient- and family-centred care for young adults living with chronic disease and their family members: A systematic review. Int J Integr Care. 2018;18(2):14.
57. Lauderdale-Littin S, Blacher J. Young adults with severe intellectual disability: Culture, parent, and sibling impact. Journal of Intellectual & Developmental Disability. 2017;42(3):230–9.
58. McGuire S, Shanahan L. Sibling experiences in diverse family contexts. Child Development Perspectives. 2010;4(2):72–9.
59. McHale SM, Updegraff KA, Shanahan L, Crouter AC, Killoren SE. Siblings' differential treatment in Mexican American families. Journal of Marriage and Family. 2005;67(5):1259–74.
60. Updegraff KA, McHale SM, Whiteman SD, Thayer SM, Delgado MY. Adolescent sibling relationships in Mexican American families: Exploring the role of familism. Journal of Family Psychology. 2005;19(4):512–22.
61. Guan SSA, Fuligni AJ. Changes in parent, sibling, and peer support during the transition to young adulthood. Journal of Research on Adolescence. 2016;26(2):286–99.
62. Walsh S, Shulman S, Bar-On Z, Tsur A. The role of parentification and family climate in adaptation among immigrant adolescents in Israel. Journal of Research on Adolescence. 2006;16(2):321–50.

CHAPTER EIGHT

Discussion

The overall aim of this dissertation was to deepen the understanding of experiences of Canadian siblings of youth with disabilities during the transition to adulthood and to synthesize the characteristics of existing programs and resources to support them in their roles. In this chapter, I discuss this dissertation addressed knowledge gaps, strengthened existing literature, and identified novel areas for future direction. A summary of the implications of these findings for families, practice, and future research is provided. Then, I share a personal reflection of my partnership with the Sibling Youth Advisory Council who have been integral to my dissertation since the very beginning. Based on my personal experiences, I provide recommendations for future researchers who are interested in establishing and facilitating an advisory committee with family partners, as well as for young adults who are considering to be engaged in research. I conclude this chapter with my next steps as an emerging patient-oriented researcher.

Key contributions of this dissertation

The overall aims of my dissertation were to develop a deeper understanding about the experiences of siblings of youth with a neurodisability during the transition to adulthood and to synthesize resources for siblings of individuals with disabilities to support them in their roles. My dissertation is comprised of three scholarly works (i.e., review and qualitative document analysis in Chapter 2; scoping review in Chapters 3 and 4; and BEST SIBS Study in Chapters 5, 6, and 7) to address the overall aims of my dissertation and identified knowledge gaps. Findings from this thesis offers contributions as a knowledge synthesis of existing literature, knowledge generation to develop a deeper understanding of the experiences of TD siblings, and knowledge to guide development of resources to support TD siblings in their roles to their sibling with a neurodisability. A summary of how my dissertation addresses each aim and contributes to existing literature is provided below.

Knowledge synthesis of existing resources and programs in neurodisability for typically developing siblings

The first aim of my dissertation was to synthesize resources for siblings of individuals with disabilities. To achieve this aim, two reviews were conducted: a review and qualitative document analysis of resources from organizations, treatment centres, and children's hospitals that are part of Children's Healthcare Canada (Chapter 2 (1)) and a scoping review to identify characteristics and outcomes of programs to prepare siblings in their future roles (Chapter 3 described the protocol (2) and Chapter 4 described the results of the scoping review).

The first review (Chapter 2 (1)) synthesized, identified, and categorized resources that are available to siblings of individuals with a chronic health condition from Children's Healthcare Canada. As a national association, Children's Healthcare Canada aims to implement a systems approach to improve the health and care delivered to children (3). The members of Children's Healthcare Canada include organizations, treatment centres, and children's hospitals who deliver health services to children and youth (3). The findings from this first review identified that most resources were focused on general information, such as booklets and books, podcasts, programs and workshops, and news articles. However, there was limited information and resources specific to healthcare management. In a Canadian needs assessment survey that was conducted across Canada (4), siblings across all ages preferred to receive information online. Based on previous experiences that the SibYAC had with searching for resources online, they described how resources can be difficult to find and websites are challenging to navigate. This review is the first step to offer a synthesis of available online resources for TD siblings in Canada.

Important topics raised in the blogs and interviews specifically by the siblings were their development of knowledge and skills for healthcare management, as well as the development of their role and identity. Siblings can provide different types of supports, including concrete or tangible support, emotional support, advice support, and esteem support (5,6), and this review further identified that siblings described how they needed to have knowledge and skills in order to support their sibling with a chronic health condition with healthcare management. For example, they needed to understand the health condition as well as the different types of treatments and services that was being provided to their sibling with a chronic health condition. This knowledge may also influence the types of responsibilities that siblings have in the future. Some of these responsibilities may be related to caregiving, which has been defined to include physical care, material and psychological support, and concern about care (7). While the blogs and interviews from siblings did not explicitly mention caregiving, the knowledge and skills that siblings require for healthcare management might also be transferable to other responsibilities such as possible caregiving in the future. Alongside the development of their knowledge and skills for healthcare management, siblings also described the development of their roles and personal identity. Many siblings described that they were experiencing their own transitions, that typically occurred during emerging adulthood (8), including moving away for post-secondary education or employment. Siblings needed time to learn knowledge about their sibling with a disability, form skills to support their sibling with a disability, and develop their identity of who they are and how they can support their sibling with a disability in the future.

Siblings also described in blogs and interviews about the importance of advocacy, including about raising awareness of the health condition of their sibling and their roles as a sibling. For example, siblings described how they explained about the health condition of their siblings to their classmates. Siblings further described the important roles that they have to their sibling with a chronic health condition, and they needed to advocate about these roles to different individuals. For example, siblings identified that employers should also recognize how siblings may need to take time of work to care for their sibling with a chronic health condition. To understand the patterns and motivations of advocacy, the context and intended recipient(s) of the advocacy action would need to be identified (9). Case advocacy refers to the advocacy for a specific individual, for example, raising awareness about the specific health condition of the sibling as described in the review (1) or advocating for specific services for an individual with a disability. Systematic advocacy occurs when there is advocacy for changes for all individuals, for example, when all TD siblings are prepared to advocate for changes in the delivery of services to individuals with a disability (10). From the literature, a qualitative study discussed the experiences of siblings in advocacy initiatives for their sibling with an intellectual and developmental disability (9). Siblings often advocated out of love and obligation to their sibling with a disability and to ensure that their sibling receives the required services (9). This review contributes to the existing literature that in addition to research studies, siblings are discussing the importance of advocacy in blogs and interviews.

The second review (Chapter 3 outlines the protocol (2) and Chapter 4 describes the findings) further builds on the important topic about the knowledge and skills that siblings require in order to provide support to their sibling with a neurodisability. This second review focused on synthesizing and mapping the characteristics and outcomes of programs designed to prepare typically developing (TD) siblings in their future roles to support their sibling with a neurodisability. These programs focused on two main areas: providing knowledge and/or acquisition of skills for the TD sibling, such as information about disabilities or development of coping skills; or empowerment by training TD siblings to teach skills to their sibling with a neurodisability, such as behavioural modification skills or social communication skills. This review also synthesized information about how families are involved in these programs.

This second review also illustrates that the historical trend of increasing sibling programs has primarily been observed in high-income countries. Among the 58 included studies, the majority of studies about sibling programs were conducted in high-income countries, including Australia, Canada, Greece, Ireland, Norway, Sweden, Taiwan, the United Kingdom, and the United States. There was only one study (11) conducted in Turkey, an upper-middle-income country, and one study (12) conducted in Cambodia, a lower-middle-income country. The classification of these countries as high-, middle-, or low-income country is based on data from the World Bank (13). The World Bank is an organization that provides support with policy recommendations, research, and technical assistance to countries (14). This second review identified that across countries, research teams were able to learn, adapt and incorporate new components into sibling programs over time. For example, there were two sibling programs that were each conducted in 2002 and 2020 (15,16) used components from sibling support workshops that were previously conducted and first published in 1994 (17). Cambodia also published a study about the effectiveness of a family intervention that included components for both parents and siblings (12). The research team in Cambodia conducted this study because there were positive results for both parents and siblings of an individual with a neurodisability when the intervention was provided in Norway (18). This second highlights how sibling programs evolve over time based on learnings from previous programs, and that international teams can learn about the successes and challenges of delivering sibling programs. While there are limited publications about sibling programs that are offered in low- and middle-income countries, there may be programs that are offered but not yet disseminated internationally.

Furthermore, this second review emphasized the importance of the family context, by describing the involvement of the family in programs and also the family home as the setting to conduct the programs. Some programs included components to have parents involved in the sessions, which was dependent on the age of the sibling. For example, parents completed questionnaires or observed their children's behaviour when the sibling participant was approximately 3-17 years old, and parents were involved in the programs as participants to receive training or information when the sibling participant was approximately 8 to 21 years old. For programs that trained the TD siblings to teach specific behavioural skills to the sibling with a disability, the parents were involved by explaining the skill first to the TD sibling (19). Some programs included integrated components, in which parents and the TD siblings could interact, and TD siblings could share their personal experiences and challenges (12,18,20). These integrated sessions could provide opportunities for parents and TD siblings to have conversations, in order to understand each other's perspectives, roles, and goals in the family. In addition to the involvement of the family in programs, most programs that focused on training the TD sibling took place in the family home environment. The family home may be a place where both the TD sibling and sibling with a neurodisability may feel most comfortable in.

Knowledge generation of sibling experiences for transitions in neurodisability

The second overall aim of my dissertation was to generate a deeper understanding about the experiences of siblings of youth with a neurodisability during the transition to adulthood. To achieve this aim, qualitative case study titled: BrothErs and Sisters involvement in healthcare TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study was conducted. The protocol was published before data analysis was completed (21). The focus of Chapter 6 is to provide a description of the different types of roles that siblings had to their sibling with a neurodisability, and similar descriptions of these roles were previously identified in a survey among adult siblings of individuals with intellectual and developmental disabilities (22). In contrast to the findings from this BEST SIBS Study, recent studies identified that siblings may not want a role with their sibling with a disability and were not expected by the family to assume caregiving responsibilities (22,23). This contrast in findings may be due to the sample size, as siblings who participated in the BEST SIBS Study may have an interest in reflecting and sharing their experiences about their roles to their sibling with a neurodisability.

Sibling relationships are complex, and there are multiple factors that can influence the roles that TD siblings have to their sibling with a neurodisability (24). Chapter 6 further contributed knowledge about the emotional level of responsibility that accompanies certain roles. The roles that siblings had may grow over time, in which as children, TD siblings are friends with their sibling with a neurodisability. As both siblings grow up, TD siblings may have additional responsibilities with the roles of being a role model/mentor, protector, advocate, or supporter to their sibling with a neurodisability. In these roles, TD siblings were carrying the responsibility to protect their sibling with a neurodisability; for example, TD siblings make sure that they are available for support, while not expressing their negative emotions or stress or explaining about the neurodisability to their friends and extended family. As many siblings have described in the literature, there is a spectrum of emotions that they experience ranging from guilt and annoyance to joy and affection (25,26). This study further describes how these emotions are attached to certain roles that siblings have. The highest level of emotional responsibility comes with being a caregiver, and some sibling participants in this study described that they gradually took on responsibilities such as taking care of their sibling with a neurodisability when their parents were not available, providing assistance with daily activities such as personal hygiene, or taking their sibling with a neurodisability to healthcare appointments. This increased emotional toll associated with an anticipated caregiver role has been described by TD siblings (23,27,28). Even in sibling relations that were not considered to be close, TD siblings have often described that they were willing to take on the caregiver role to their sibling with a neurodisability when their parents are not longer able (27).

Chapter 6 also advanced knowledge about how siblings processed and decided on the roles that they have to their sibling with a neurodisability. There were four factors that influenced how siblings processed the roles that they had to their sibling with a neurodisability, which included: acquiring knowledge, preparing plans, making adjustments, and seeking supports. The acquisition of knowledge was previously identified in the two reviews of my dissertation (1,2). This study identified that the additional steps of preparing plans and making adjustments was a continuous process, in which siblings needed to be ready for changes that took place including when their sibling transitions after graduating from high school and transitions to the adult healthcare system. Alongside the transitions of their sibling with a neurodisability, siblings are also experiencing their own transitions of graduating from high school, pursuing postsecondary education, and/or finding employment (8). Sibling participants in this study also identified the importance of seeking supports for their mental health and well-being, which is a key factor in the decision-making process of their selected roles. Mental health supports can be informal from various individuals who they trust, such as their parents/caregivers and friends including those with or without a sibling with a neurodisability. Siblings are continuously processing these four factors, which can change the roles that they have to their sibling with a neurodisability.

In Chapter 6, the three factors of personal identity, values, and experiences, collectively known as intrapersonal characteristics, were described that influenced the roles that sibling participants wanted to have to their sibling with a neurodisability. Age and gender were considered to be a part of the personal identity of sibling participants. These intrapersonal characteristics are variables similar to the literature in how siblings choose to have the role of a caregiver (29–31). Further, adult siblings are expected to provide future care if they are the only typically developing sibling in the family, identified as female, lived nearby, and had a close emotional bond to their sibling with an intellectual or developmental disability (29,30). In addition to intrapersonal characteristics, this study also identified the influence of values and personal experiences. Personal, cultural, and family values influenced the extent to which siblings felt the need to have certain roles to their sibling with a neurodisability. Some siblings also shared that they had a disability themselves, and they wanted to share their own experience with the healthcare system to support their sibling with a neurodisability.

Findings from Chapter 7 provides further knowledge about the identity that TD siblings form based on their relationships with their sibling with a neurodisability and family. The evolvement of different sibling relationships occurred alongside the TD siblings' own development of their personal identity. The influence of the sibling relationship on TD siblings can be considered as "disability by association", in which TD siblings and their family are impacted by the disability in different ways (32,33). This study provides a deeper understanding of the phenomenon of “disability by association”. Within the family, TD siblings are often “filling in the gap” as they want to provide the best support that they can. They had to process how their role best fits within the family as the puzzle piece. The formation of their identity as a sibling to an individual with a neurodisability in the family developed over time. As part of their identity, some siblings developed problem-solving skills as they wanted to avoid overburdening their parents with additional challenges that they were facing. In the literature, TD siblings of individuals with a neurodisability may develop strengths, such as open-mindedness, kindness, love, leadership, and gratitude (34) in addition to problem-solving skills as identified in this study. In addition to their personal development, the relationship that TD siblings had with their sibling with a neurodisability also evolved over time. For example, the TD siblings learned to have a mature and open-minded perspective with their sibling with a neurodisability. As the TD siblings grew older, they recognized how to mediate and address conflicts that they used to have with their sibling with a neurodisability. Having a sibling with a neurodisability provided benefits to TD siblings to develop these positive traits, such as being open-minded and patient, which have been described by TD siblings of individuals with a disability in qualitative studies and systematic reviews (35–40). TD siblings also had to process and communicate with other TD siblings in the family. For example, if one or more siblings were moving away from home, there needed to be delegation of roles. The literature often discusses about sibling dyads (41,42), and this study further adds knowledge about the relational support that multiple TD siblings in the family may provide to each other.

Findings from Chapter 7 provide a further understanding about these family relationships have an influence on the identity formation of TD siblings. TD siblings had different types of identities that were influenced by their family relationships, which includes their sibling identity, student identity, social identity with their peers and romantic partner, and professional identity in their careers. A qualitative case study in the literature described a 39 year old woman with a 35 year old brother with Down Syndrome, and her identity that was shaped by her experiences as a sibling (43). For example, she identified the importance of finding a romantic partner who would be understanding of her brother, and that she also wanted to continue to live with her brother (43). Findings from this BEST SIBS Study further strengthens existing research, as the experiences that TD siblings have continuously shape their identity. During adolescence and emerging adulthood, TD siblings are planning for how their sibling with a neurodisability will be a part of their lives. The literature has identified that siblings may choose to move away from home for postsecondary education or careers, and their communication with their sibling with a neurodisability may be different (44). This BEST SIBS Study identified that siblings were making plans about how their sibling with a neurodisability will be a part of their current and future life stages; for example, sibling participants described their worries of who would take care of their sibling with a neurodisability and family, or if they will move back home to be near their family after completing postsecondary education. The process of making these plans can influence the choices that TD siblings make in their social identity and/or professional identity, such as the future family that they may create or the careers that they might have. Similar to the findings from the blogs and interviews identified in the review and qualitative document analysis (Chapter 2, (1)), siblings in the BEST SIBS Study also described how their sibling relationship influenced them to enter professional careers that could help others with a neurodisability. Existing research also has similar findings, in which TD siblings may choose to enter professional careers such as the disability field or professions that could help others (44,45).

Overall, the findings presented in Chapter 7 highlights that the experiences that TD siblings have with their sibling with a neurodisability, other TD siblings, and parents/caregivers influenced the different types of identity that they have, including their social and professional identity.

Knowledge to guide development of support resources for typically developing siblings

My dissertation was focused on the knowledge creation cycle, including knowledge inquiry and knowledge synthesis of the Knowledge-to-Action Framework (46). This dissertation synthesized knowledge of sibling resources, as well as generated knowledge about the experiences of siblings in their roles and identity formation. This dissertation can inform future work about: 1) knowledge transfer; and 2) applying knowledge to guide the development of resources and interventions.

The first step of knowledge translation is the transfer of knowledge, in which information is shared to key stakeholders (46). The knowledge synthesized from the two review papers (Chapter 2 (1), Chapter 3 outlines the protocol (2) and Chapter 4 describes the findings) can be shared with target audiences, including siblings and families of youth with a disability, professionals who work with these families, healthcare organizations and policymakers. Discussions have been held with the SibYAC to share the synthesized knowledge. Resources and programs identified in Chapters 2 and 4, respectively, can be shared as website links to the organizations as a Sibling Knowledge Hub on a website. For example, information about the BEST SIBS Study (Chapters 5) is provided on the website hosted through the CanChild Centre for Childhood Disability Research (47), and an additional website tab can be created for the Sibling Knowledge hub. Previous knowledge hubs have been created for the Health Hub in Transition in Canada (48) and the F-words for Child Development Knowledge Hub (49). Initial conversations have also begun about how a Sibling Knowledge Hub could be disseminated by childhood disability organizations

A remaining gap in my dissertation is the lack of resources for young adult siblings of youth with a disability who are transitioning from pediatric to adult healthcare. To address this gap, a potential project is to co-create and evaluate a knowledge product/tool, tentatively titled as a 'Sibling Kit' with multiple stakeholders including siblings, parents/caregivers, healthcare providers, and policymakers. The involvement of multiple stakeholders in the co-creation process helps to ensure that the tool is applicable to a broad target audience. For example, a parent and youth resource is available, called Keeping It Together (KIT) comprised of worksheets to learn how to provide information about themselves, receive information from others, and organize information during transition to adulthood (50,51). While there is a resource sibling kit titled, the SibKit, that provides tools and tips for siblings of children with disabilities (52), a specific resource can be adapted or co-created to support TD siblings in their roles during healthcare transition of their sibling with a neurodisability.

Programs can be provided to support siblings and the whole family. A family systems approach illustrates that there is a bidirectional relationship between siblings and the family (53). Programs can be developed that allows all family members to participate with tailored sessions for each family member. As described in the second review (Chapter 4), family programs can be provided with separate components for parents and TD siblings with an integrated session for parents and TD siblings to interact with each other (12,18,20). TD siblings require knowledge and skills to support their sibling with a disability (as identified in Chapters 2 and 4). These skills can include skills that TD siblings develop for themselves, such as coping skills, as well as skills that can be applied to their sibling with a neurodisability, such as how to communicate with their sibling with a disability (as described in Chapter 4). This knowledge about information to include in the sibling program sessions further adds to the literature. Programs can be offered to families when their children are at different developmental stages.

The literature describes programs that are offered to parents for children that are focused on addressing sibling conflict (24,54,55). For example, a program describes mediation strategies that were taught to parents, including reasoning, discussing emotions and understanding motivations of the children to address conflicts (54). While the program was developed for typically developing children, similar strategies could be taught to parents to support the development of the sibling relationship between their child with a disability and typically developing child. During adolescence, the family is often focused on preparing for the transition to adulthood of their child (56,57). Existing programs to support families during transition could be enhanced, in which the goals and roles of each member of the family including the sibling(s) can be discussed. For example, residential immersive life skills programs are designed for youth with disabilities to prepare for adulthood (58), and there could be a component in which siblings could be involved in this transition process.

Implications

My dissertation has overall implications for stakeholders, including researchers, families, and healthcare providers. These implications have also been discussed with the SibYAC, in which they wanted to ensure that the scholarly works in my dissertation brought awareness to the important and valuable roles of siblings of youth with a disability. The implications can be categorized into the levels of the bioecological systems as described Bronfenbrenner (59):

microsystem with the family; and the macrosystem with the delivery of healthcare services and creation of government policies.

Microsystem at the level of the family. In my dissertation, there was an identified need about the importance of having family conversations to discuss future plans when the youth with a disability was preparing for transition, including the transitions of graduating from high school and from pediatric to adult healthcare. The extent to which families discuss about the roles of siblings in the future can vary (60,61); when there are discussions, there is often a lack of concrete actions of the steps that can help siblings to prepare for their future roles (62,63). Conversations, including ones within the family and with healthcare providers, could consider the whole family in the care of the youth with a disability. The family context is critical to the health of an individual, as illustrated by the International Classification of Functioning, Disability, and Health (ICF) framework and translated into the 'F-words' of fitness, function, friendships, family factors, fun, and future. A positive family environment where family members, including siblings and youth with a disability, can support each other can then in turn, promote optimal health for the youth with a disability and family. Family conversations can be ongoing and fluid, as each family adapts to changing roles and level of readiness for transition.

Macrosystem at the level of healthcare provision. There should be recognition about the roles of siblings as part of the whole family unit in the delivery of healthcare services. During the preparation of transition for youth with a neurodisability, professionals could have conversations that incorporates the perspectives of all family members. There can be questions to ask families about the roles that siblings might have. For example, an article with a hypothetical case study described how siblings of students with disabilities can be included in discussions between the family and school professionals during the planning of transition, including postsecondary education, employment or community participation (64). School professionals can work with siblings to identify their strengths and preferences, and empower them to be involved in transition planning (64). Similarly, during the discussions to prepare youth with disabilities during the transition to adult healthcare, professionals can have a conversation with the family about how TD siblings can be involved. Findings from this study further supports how some siblings either have an implicit role or choose to be involved in their roles. Open conversations help ensure that all members of the family, including TD siblings, have clarity about their roles.

Macrosystem at the level of government policies. There should be awareness and establishment of policies to recognize the roles that TD siblings might have to their sibling with a disability. In 2018, there were 7.8 million Canadians ages 15 or older which is 25% of the population who were caregivers to their family member or friend with a long-term condition, a physical or mental disability or aging-related problems (65). Canadian organizations have advocated for government funding of supports for informal caregivers of an individual (66–69). These supports can include community supports that consider the well-being of caregivers such as informal groups where caregivers can connect with each other, financial assistance such as a tax credit to cover medical and disability-related expenses, and rights to request accommodations at work such as offering flexible hours or time off from work when needed (66). Yet, there needs to be recognition that informal caregivers also include TD siblings of an individual with a disability. For example, in the BEST SIBS Study (Chapter 6), some siblings including those as young as age 14 were processing how they may become future carers to their sibling with a disability. One of the findings in the review and qualitative document analysis (Chapter 2, (1)) identified that adult siblings who were carers to their sibling with a disability were often not recognized for this important role at work. Other co-workers may take time away from work to care for a child, while adult siblings were not recognized for taking time off to care for their sibling with a disability. Sibling carers to their sibling with a disability can range in age, from young carers under the age of 18 or adult carers (70,71). A first step could include the development of government strategies and policies with explicit statements that recognize and acknowledge the important role of sibling carers. For example, in Australia, the National Carer Strategy launched in 2010 that outlined a 10-year plan to improve the lives of individuals with disabilities, their families including siblings, and carers (72). The social inclusion statement included in this plan recognized different types of carers, including older and young carers, as well as those from culturally and linguistically diverse backgrounds (72). A similar strategy could be developed in Canada that acknowledges the role of sibling carers, and this movement towards the national awareness of siblings has been a focus for the Siblings Canada initiative (73). This initiative is a part of the Canadian Centre for Caregiving Excellence that focuses on making an impact about the role of siblings of an individual with a disability through research, knowledge and policies (73). Findings from my dissertation strengthens the need to raise awareness of sibling roles, including being carers, in government policies and strategies in Canada. These types of government strategies can then lead to interventions and programs to support sibling carers, which could be tailored based on their age.

Strengths and limitations of this dissertation

The strengths and limitations are discussed within each manuscript, and I reflect about the strengths and limitations across all scholarly works in this dissertation.

First, the scholarly works further strengthened existing literature by synthesizing resources for siblings of an individual with a disability and developing a deeper understanding about the experiences that TD siblings have in their roles and responsibilities and how these experiences influenced their identity formation. Findings from the two reviews, the review and qualitative document analysis (described in Chapter 2 (1)) and the scoping review (with the protocol described in Chapter 3 (2) and results presented in Chapter 4) complemented each other. The review and qualitative document analysis identified and synthesized existing online resources, as well as key topics that siblings and families were sharing in blogs and interviews. The scoping review identified the historical trend about the types of sibling programs that were available, which primarily focused on knowledge acquisition and skill development for the TD siblings themselves and empowerment of the TD siblings to teach skills to their sibling with a neurodisability. Novel findings about the experiences of TD siblings in families of individuals with a neurodisability were presented with the BEST SIBS Study (with the protocol described in Chapter 5 (21), and findings presented in Chapters 6 and 7). The scholarly works offers future directions in that programs could be provided to TD siblings that could support them in their current and future roles.

Second, this dissertation employed a descriptive case study methodology by Yin for the BEST SIBS Study (with the protocol described in Chapter 5 (21), and findings presented in Chapters 6 and 7). A descriptive case study methodology provided guidance with the use of theories as a conceptual framework with propositions (74). With the use of Yin's methodology, I reflected about how I can reconcile the different positionalities. Yin positions himself as a post-positivist while I position myself as a social constructivist. In recent writings, Yin acknowledges that a social constructivist and interpretivist approach can be valuable in a case study methodology (75,76). In addition, a descriptive case methodology offered guidance in the use of theories to design, conduct, and analyze the data. This dissertation provides knowledge about how theories can be applied in research. The methodology of a case study (74) provided flexibility for two theories to be used as a guide for design, conduct, and analyze data for the BEST SIBS Study (Chapters 5-7). The two theories that were applied in the BEST SIBS Study included the transition theory developed by Meleis and colleagues (77) and the bioecological systems theory developed by Bronfenbrenner (59). The two theories were beneficial in formulating the conceptual framework and propositions to ensure that there were boundaries about the scope and feasibility of completing the study (78,79). Furthermore, the bioecological systems theory provided guidance to identify a clear focus for each chapter about the findings for the BEST SIBS Study, in which Chapter 6 focused on the microsystem about the roles and responsibilities that TD siblings have in their family and Chapter 7 focused on the mesosystem about the influence of the interactions between family relationships on the identity formation of TD siblings. The focus on the experiences that TD siblings have in their microsystem and mesosystem strengthened existing literature about the importance of considering the family context in research and healthcare systems (80,81). The literature has also described that the environment, including factors in the microsystem and mesosystem, can have an impact on the development of an individual (81). Similarly, findings from the BEST SIBS Study highlights that the experiences that TD siblings have in their microsystem and mesosystem will continue to change over time during the developmental stages of childhood, adolescence, and emerging adulthood.

Third, a descriptive case study methodology offers flexibility in the analysis of the data (82). I sought additional guidance to analyze the data by using reflexive thematic analysis (83). The steps outlined in reflexive thematic analysis offered guidance to develop codes and categories in an iterative process with data collection and data analysis. Further, peer debriefing was a methodological strength that was employed as a technique in data analysis across the three scholarly works. I had discussions with a multidisciplinary team of researchers with expertise in cognitive psychology, education, nursing, occupational therapy, physiatry, rehabilitation, patient-oriented research, and lived experiences. Peer debriefing can be used to ensure credibility of the study findings in which there is a discussion about preliminary themes, alternative ideas or approaches, and rename themes (84,85). This technique of peer debriefing has been used in multiple qualitative research studies (86–90), and researchers can find support and compassion about sensitive information shared by participants in qualitative studies (87). There were multiple levels of peer debriefing, in which SibYAC members were asked about the level of engagement that they would like to have during data analysis using the Involvement Matrix (91). For example, the SibYAC member could have the role of being a co-thinker in which they are solicited for advice, an advisor in which they provide both solicited and unsolicited advice, or a partner in which they collaborated throughout the process of data analysis. The input and perspectives that SibYAC members had on the data and preliminary themes further contributed to the rich and deep level of analysis. For example, the SibYAC were co-authors for Chapter 2 (1) in which they reviewed the results and provided their reflections, were advisors for Chapter 4 to share their thoughts about the implications of the findings, and were partners in the BEST SIBS Study in which they shared their perspectives on the preliminary themes during the data analysis phase. Based on individual conversations with each SibYAC member, some chose to be engaged with the manuscripts of the results of the BEST SIBS Study (Chapters 6 and 7) as co-authors, while others preferred to be acknowledged in which they provided general thoughts on the paper and would like to be updated about the status. I came to understand how partners can be engaged in manuscripts in different ways and listed as co-authors or acknowledged depending on their role (92). These different levels of engagement from the SibYAC provided me with valuable input to understand the data, including the meaning and significance of the study findings, implications, and future directions.

Fourth, I engaged in reflexivity as an ongoing process that allowed me to consider how my identity and background has an impact on the research process across all scholarly works as part of my doctoral program of research (93,94). I wrote reflexive memos during the conduct of all scholarly works to document my prior knowledge from reading the literature about sibling relationships, change in thoughts while analyzing the data, and ideas that could be incorporated based on the perspectives from the SibYAC. These reflexive memos were also beneficial to identify how the findings of each scholarly work connected to each other as part of a broader doctoral program of research, and the role that I had in shaping and identifying the importance of each scholarly work. During the data analysis of all scholarly works in my dissertation, reflexivity allowed me to be aware and better understand the data and presentation of the finding, as well as how the findings of each scholarly work were connected and can build on each other in this dissertation (95). I consistently asked myself questions including “What is occurring in the data?”, “How come this finding was interesting to make note of?”, “How did perspectives from the SibYAC and multidisciplinary team influence how I viewed the data?”, or “How might the findings of this scholarly work relate to the findings of the other scholarly works?”. Ongoing reflexivity allowed me to conduct data analysis in an iterative process to develop key themes and findings that were comprehensive with detailed information. As my thought process continued to evolve, I shared this process with the SibYAC and asked about their perspectives or if they had questions about my analysis. The iterative process of data analysis with the full team of both my supervisory committee and the SibYAC continually challenged my thoughts in how I viewed the data, until we came to a consensus on the themes and key messages of the data.

Fifth, in addition to reflexive memos, I kept an audit trail with a record of my notes about key decisions that were made. An audit trail can be considered as a document of methodological and analytic decisions that are made in the research study (96,97). After the protocols were written for the scoping review (Chapter 3, (2)) and BEST SIBS Study (Chapter 5, (21)), I kept a working document about changes that were made to the methods and thoughts about the progression of the analysis (97). For example, there needed to be more clarity in the information that was being extracted for the outcomes of sibling programs in the scoping review, and I made a change to the data extraction sheet to identify and distinguish outcomes for the sibling with and without a neurodisability. The analysis of the BEST SIBS Study also led to the presentation of findings as two separate papers with a distinct focus (Chapters 6 and 7), and I documented my thought process of how I came to identify the focus of each paper.

There were several limitations of my dissertation, which could potentially be explored in future research. First, the focus of my dissertation was on TD siblings of youth with a disability who grow up with their parents/caregivers in a family home environment. The experiences of siblings from my dissertation may differ from the experiences of those who grow up in an institution or foster care (98–100), in which they may be living separately from their sibling(s) and/or parents/caregivers.

A second limitation is that my dissertation did not focus on the influence of the cultural context on the development of sibling relationships within the family. While some sibling participants in the BEST SIBS Study (Chapters 6 and 7) shared how their cultural values influenced the roles that they had within the family, I recognize that it was a limitation that demographic information about race and ethnicity was not collected. Future studies can be conducted using intersectionality theory to understand, acknowledge and critically examine how multiple socially-constructed identities (e.g., race, gender, sexual orientation, class) shape the experiences of individuals within systems (101,102). Intersectionality theory describes the assumption that individuals have multiple identities; individuals experience power or oppression within each identity; and these identities are formed within a socio-cultural context (103). In a scoping review, cultural values have been found to influence the experiences of TD siblings of individuals with intellectual and developmental disabilities including their understanding about disability, expression of emotions, and caregiving responsibilities (104). Additional research is needed using intersectionality theory to understand how different socially-constructed identities, including culture, race, and ethnicity, affects the relationships of relationships of siblings with and without a disability

A third limitation is that my dissertation focused on the Canadian context with only articles written in English included in the two reviews (Chapters 2 and 4), as well as interviews conducted in English (Chapters 6 and 7). Future studies can be conducted to understand the similarities in the experiences, roles, and responsibilities of TD siblings of youth with a neurodisability in other countries.

Personal reflection on the partnership with the Sibling Youth Advisory Council

The partnership with the Sibling Youth Advisory Council (SibYAC) was critical towards shaping this dissertation. A patient-oriented research approach was valuable to ensure that this research topic was relevant, meaningful and applicable to siblings with lived experiences. The Canadian Institutes of Health Research defines the term of ‘patients’ broadly to include individuals with personal experience with health, including their family and friends. With the increasing movement towards patient engagement in health research, ongoing reflections about the process of engagement could lead to important recommendations to continue authentic, meaningful partnerships (105). Previous doctoral students have reflected about their experiences with patient partners (106,107). In this section, I provide context about how my partnership with the SibYAC was established using key principles of patient engagement. Then, I describe how this partnership impacted three key areas: the research process, members of the SibYAC, and myself as a researcher. I conclude with lessons learned and recommendations for other doctoral students and researchers interested in engaging with patient partners in research.

Context to establish the Sibling Youth Advisory Council

The SibYAC was formed in October 2018, and it was important to keep building and sustaining our partnership together. I facilitated our partnership by having individual check-in meetings starting in January 2020 to ask each SibYAC member about their motivations for joining the SibYAC, their personal goals, and group goals. It was by learning these goals from the SibYAC, I embodied the role of a facilitator to support each SibYAC member with their individual goals but also our group goals. After these individual check-in meetings, I shared a summary of the goals that each SibYAC member had at a SibYAC meeting. As illustrated in Figure 1, after the individual check-in meetings, we engaged in multiple initiatives. At a recent SibYAC meeting held on February 14, 2022, each SibYAC member and I identified five moments that stood out to us. These five moments, with some moments that were similar to each other, are mapped on Figure 1. Our initiatives only began after the individual check-in meeting on January 2020; for example, we had our first academic conference, collaboration with other youth partners, and guest speaker at a group meeting.

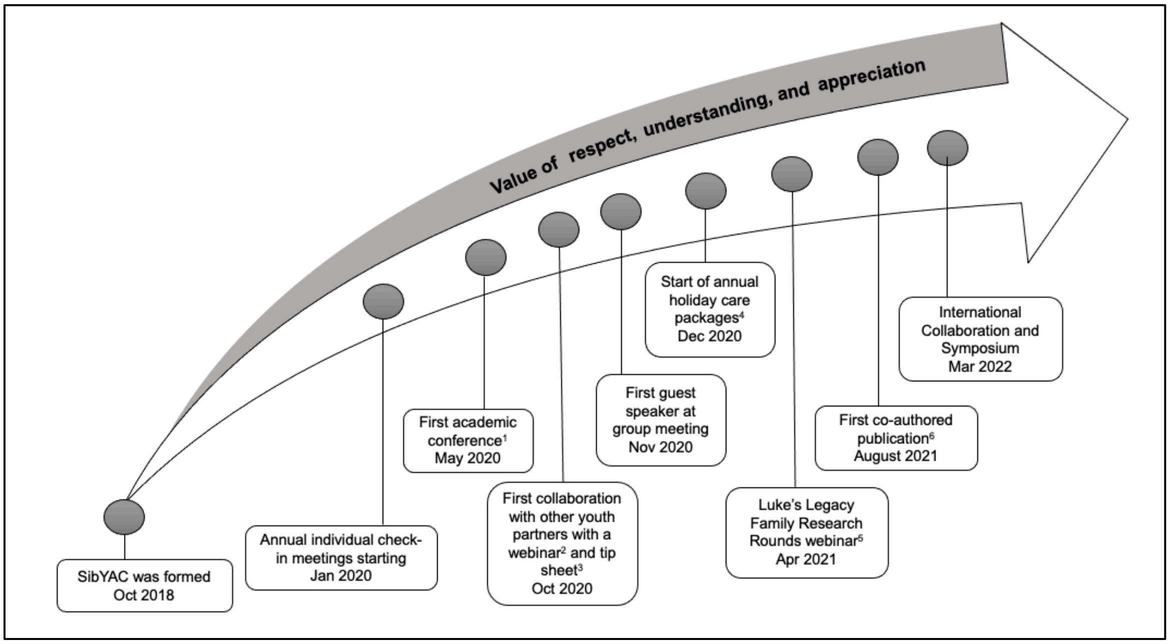

Figure 1. *Moments of significance in the partnership between the SibYAC and myself.*

In collaboration with the SibYAC members, a mutual set of goals were developed:

1. To raise awareness and advocate for the important and unique role of siblings of individuals with disabilities in order to give siblings a voice;
2. To provide support to each other as a group.

Individual SibYAC members also shared the personal outcomes they anticipated achieving through their participation which included: 1) establishing connections with the academic community; 2) disseminating study findings through conference attendance and publishing; 3) improving communication skills with respect to public speaking; and 4) creating a community with a sibling support network to share stories and experiences.

To achieve these goals, an investment of time was important (as illustrated in Figure 1). Time was also needed to continue to build and continue my partnership with the SibYAC. The building of this partnership included an application of the principles of patient engagement outlined by the Canadian Institutes of Health Research in our partnership, which included: inclusiveness, support, mutual respect, and co-build (108). Supplementary Table 1 outlines how each of these values were displayed by the SibYAC and myself.

Engagement of the Sibling Youth Advisory Council in this dissertation

The SibYAC were invited to be engaged in all research phases of my dissertation including: 1) preparation; 2) execution; and 3) implementation. I describe the overall impact of the SibYAC on the research process below. Details about the involvement of the SibYAC in each research phase of this dissertation is provided in Table 1.

Preparation. Their perspectives had a significant influence on the research topic, in which they identified that prior to the development of an intervention for siblings, it was important to raise awareness about the important multi-faceted roles that siblings had. They identified that the broad topic of 'sibling experiences' was important to understand. Within this broad topic of my dissertation, the SibYAC were then involved with discussing the purpose of each manuscript in this dissertation. To conclude my overall dissertation, the SibYAC further discussed how they wanted to contribute Chapter 8 with the Discussion.

Execution. The SibYAC were most actively engaged in the qualitative BEST SIBS Study described with the protocol described in Chapter 5 (21), and results presented in Chapters 6 and 7. They were involved with the co-creation of recruitment materials available on our study website (47), and were invested in sharing the recruitment materials in their personal networks on social media. In addition to the collaboration from the SibYAC to conduct the study, the SibYAC were invited to be involved in all scholarly manuscripts. This involvement included an invitation to be co-authors or acknowledged in the manuscripts, and a discussion about the meaning of authorship was discussed with the SibYAC using guidance from the literature about how to engage patient partners in authorship roles (92). Their engagement in each scholarly paper is described as follows. In Chapter 2 with the review and qualitative document analysis (1), the SibYAC shared their reflections about the importance of synthesizing resources and highlight what the findings mean to them. These reflections provide guidance for future directions about the resources that are needed to support siblings. In Chapters 3 (2) and 4 about the scoping review of programs to support siblings in their roles, the SibYAC preferred to be kept updated about the synthesized results. In Chapters 6 and 7 about the results of the qualitative BEST SIBS Study, the SibYAC have been involved in ongoing discussions to debrief about the results.

Implementation. The SibYAC identified their interest in co-presenting at conferences and webinars to share their partnership experiences, as well as their engagement in the work presented in this dissertation. As our next steps, the SibYAC and I have had discussions about how to share this dissertation work as lay summaries on our study project website (47). To continue our partnership and knowledge translation activities, the SibYAC and I have also discussed plans to apply to grants for funding. These steps towards knowledge translation activities are ongoing and will continue to be discussed beyond my doctoral program of research.

Table 1. Involvement of the SibYAC in each dissertation phase.

Research phase	Description of involvement
Preparation	- Identification of topics of importance. - Development of the research question. - Contribution to the study design. o For example: informing the key outcomes for data extraction in Chapters 3 and 4; discussing the appropriateness for the use of photo elicitation and graphic elicitation of relational maps in Chapters 5, 6, and 7.

Execution	- Participant recruitment in Chapters 5, 6, 7. - Interpretation of study findings for Chapters 2, 4, 6, and 7 based on their lived experiences and expertise as a method of data analyst triangulation and peer debriefing.
Implementation	- Dissemination of partnership experience and preliminary study findings at conference co-presentations and webinar. - Sharing of our partnership experience and research outputs on our project website; for example, the publications with Chapters 2, 3, and 5 (109).

Preliminary evaluation of our partnership

To conduct a preliminary evaluation of my partnership with the SibYAC to date, I used the Public and Patient Engagement Evaluation Tool (PPEET) at two timepoints: 1) design of the BEST SIBS Study (December 2020 – January 2021) and 2) implementation activities, including sharing our partnership experiences to conduct the BEST SIBS study at conferences and webinars (March 2022). I provided SibYAC partners with the option to complete the PPEET prior to the individual check-in meetings. The questions from the PPEET may help SibYAC members to reflect on different activities and consider feedback that they would like to provide about our partnership. The PPEET was an option for SibYAC members to provide feedback anonymously on the following aspects of our partnership: A) communication and supports for participation; B) sharing their views and perspectives; C) their impacts and influence on the engagement initiative; and D) final thoughts (for example, their satisfaction with the engagement initiative). There were six members of the SibYAC and about four active members at any time of my dissertation. I asked the SibYAC to consider the type of feedback that they would like to provide and avoid providing information that could identify themselves.

Based on initial results, there were four responses provided at the first timepoint and three responses provided at the second timepoint. In both timepoints about the impact and influence of their involvement in the SibYAC has been agree or strongly agree. For the second timepoint, while all SibYAC members agree or strongly agree that they were able to express their views freely and feel that their views are heard, there could be further improvement to ensure that a wide range of views on discussion topics is shared and a representation of a broad range of perspectives. The diversity of additional perspectives as part of the SibYAC can help to ensure that the research is applicable and relevant to siblings with various experiences. The next steps to broaden the diversity of the SibYAC includes inviting siblings from different genders and cultural backgrounds to be a part of the SibYAC. The invitations can be shared by the research team including the SibYAC members in their personal networks, as well as by organizations focused on providing care and supports to families of children with a disability.

At the first timepoint, the SibYAC provided their open-ended responses to the strengths of our partnership including:

> "I can see myself in many of the projects, updates, and shape of the research. I have definitely had an impact."
>
> "It's been a true partnership! Also, the flexibility of the research team with the schedules of various sibling partners had led to a natural incorporation of different people's stories into different parts of the project."

At the second timepoint, the SibYAC shared that a strength included our increasing reach of more siblings of individuals with disabilities and sharing our stories with the community. Another SibYAC member highlighted that:

> "Consistent updates and necessary documentation are provided by our project lead, Linda, on a regular basis, making this a fantastic experience and easy to keep up to date, given I personally have a lot on the go in addition to the SibYAC."

While the SibYAC did not identify any challenges in our partnership, their responses in the preliminary evaluation of our partnership highlight strengths and activities to continue.

Moments of tension in our partnership

While the SibYAC did not identify challenges in our partnership, I reflect on the two key moments of tension that I felt in our partnership. These two key moments of tension can be considered as barriers to our partnership which include: 1) financial barrier, due to a lack of compensation at the start of our partnership; and 2) system barrier, with having a balance between providing sufficient time for our SibYAC partnership while moving forward with the timeline of my doctoral program.

Financial barrier. At the beginning of my partnership with the SibYAC, I did not have funding. As my partnership with the SibYAC was integral to my dissertation, I wanted to acknowledge and appreciate how I value our partnership. Compensation is one form of appreciation, as described by other patient partners in the literature (110,111). While I recognize that there are guidelines for compensation (112,113), I did not have funding at the beginning of my doctoral studies to provide compensation to my SibYAC partners. I had a conversation with the SibYAC to discuss how they felt tokens of appreciation, and I mentioned that there were funding opportunities that we could apply together that could offer ways that they could acknowledged and shown appreciation such as compensation. They provided support with testimonials about the importance of the proposed project for the funding application, as well as what our partnership meant to them. When we were successful with two grants, the CIHR Patient-Oriented Research Awards – Transition to Leadership Stream – Phase 1 and the CHILD-BRIGHT Graduate Fellowship for Patient-Oriented Research, there was an allocated budget for compensation. I asked the SibYAC about how they would like the compensation, in which they wanted a combination of an annual honorarium and social activities (e.g., holiday care packages, virtual social gatherings with an Ubereats e-gift card), which are outlined as suggestions in our

Terms of Reference document. Through conversations, I was able to learn about different forms of appreciation would be valued by the SibYAC.

System barrier. I also faced the barrier of creating a balance between providing time for the SibYAC to share their perspectives while also following the timelines of my doctoral research program. I had conversations with the SibYAC about my timelines and provided opportunities for the SibYAC to share their perspectives and feedback on my doctoral studies at different time points. For example, the SibYAC could share feedback at team meetings, manuscripts in this dissertation, and/or the final manuscripts that will be submitted for publication. I reflect on when I had to collate and synthesize all manuscripts for this dissertation by a certain deadline, and I provided information that the SibYAC could choose to provide feedback for the final manuscripts before submission for publication. These transparent conversations were helpful for the SibYAC to know that they had options about when they could provide feedback based on their schedule.

Overall, these two moments of tension have been described in a scoping review about the facilitators and barriers that other trainees have faced in research partnerships (114). Ongoing reflective conversations were helpful to discuss and address the moments of tension that I felt with the SibYAC.

Personal impact of our partnership on the Sibling Youth Advisory Council

Members of the SibYAC initiated the idea of how they would like to share their personal reflections about their experiences in this partnership in this chapter. One member reflected about the meaning of the SibYAC in their personal life, research, and the community:

> The SibYAC, for me, represents a voice and an anchor. Our research, knowledge translation, and community outreach activities are an opportunity for me to share my experiences and leverage them to inform our initiatives.

Multiple members of the SibYAC described the value of being connected to siblings with lived experiences of having a sibling with a disability, as described by a SibYAC member:

> Just as important as this, however, is our relationship with each other. I experience our dynamic as a stabilizing influence - a supportive, stimulating, and safe place as we all navigate our siblings' transitions as well as our own, particularly during the COVID-19 pandemic. I actively look forward to our monthly meetings, and being part of this growing work to help siblings like ourselves.

A factor that contributed to the positive dynamic is the role that I had as a facilitator that allowed them to have a positive experience as a member of the SibYAC. All SibYAC members felt there was recognition about having compassion and understanding with each other, and a SibYAC member described that the SibYAC was a place: “where us as siblings feel heard and valued as a true partner in research”. With these positive experiences, one SibYAC member shared how she has become an active member of other advisory councils to represent the perspectives of siblings of an individual with a disability. She recognized how she has valuable lived experiences to inform research, as demonstrated from her partnership with my dissertation.

Personal reflection of the impact of the Sibling Youth Advisory Council partnership on myself as a researcher

I am aware that the SibYAC is the first advisory council that I led with the support of my supervisory committee. I have stated with the SibYAC that I want to ensure that we have an authentic and meaningful partnership and for this reason, I am open to their feedback. During our individual check-in meetings, team meetings, and with the Public and Patient Engagement Evaluation Tool (PPEET) (115), the SibYAC did not mention challenges that they experienced.

Instead of challenges, I believe that I had a personal awareness about my partnership with the SibYAC. This ongoing process of personal awareness, also known as reflexivity, is a strength of how I facilitated the SibYAC. My engagement with reflexivity allowed me to see myself as a facilitator of the SibYAC. Characteristics of being a good facilitator include providing optimal circumstances for engagement, enabling contributions by removing external barriers, creating an environment for collaboration, having regular communication, and focusing on the practicalities of how to engage with partners (116). The SibYAC have described that as a facilitator, I have the personal traits to establish a meaningful partnership including taking the initiative to reach out to each member, being flexible, organized such as sending documents in advance, being open to listen, and explaining research in lay terms. Being reflexive allowed me to consider how to offer the most meaningful experience that I could for each SibYAC member.

The impact of my partnership with the SibYAC is the recognition of our shared experiences and we can provide support to each other. The SibYAC and I were growing as a team, not only as a research partnership, but also individually as we are all young adults (ages 21-28 years old) and learning how to navigate our different life transitions. For example, we were learning about which postsecondary programs to pursue, developing our career pathways, and moving away from home. I began to realize that I was a part of the group, and we can share about life events and challenges that we had, including our successes and fears. For example, we congratulated each other when a SibYAC member or myself graduated from a program or found a new job. The turning point when I realized that our partnership was bi-directional was when the SibYAC asked how I was doing. I realized at that point that while I was supporting the SibYAC, they also wanted to provide support to me as an individual.

Recommendations

For other doctoral students and researchers, I would offer the following recommendations based on my personal reflections of partnering with the SibYAC.

Recommendation 1: Invest in time to build the partnership.

Time is needed to build a partnership, as I have experienced through my partnership with the SibYAC. The SibYAC was formed in October 2018, and time was needed to keep building and sustaining our partnership together. This investment of time allowed me to build rapport with each SibYAC member, as well as time for members of the SibYAC to get to know each other and work together as a collective time. A part of building this rapport and establishing trust among the team was providing time for personal check-ins at the beginning of each meeting. These personal check-ins allowed SibYAC members to share about highlights and updates in their lives, as well as topics that they were interested in. Some of the topics related to the experiences of siblings, which could be an area of future exploration in research. In addition to the personal check-ins, time was needed to have individual check-in meetings with each SibYAC to discuss individual goals and collective goals. While the SibYAC was established in October 2018, our research initiatives only began after the individual check-in meeting on January 2020; for example, we had our first academic conference, collaboration with other youth partners, and guest speaker at a group meeting. There also needed to be time for the SibYAC to ask questions and learn about the research process, such as what data analysis might look like across each scholarly work in this dissertation.

Recommendation 2: Be mindful to incorporate reciprocity in the partnership.

While the SibYAC was a partner in my doctoral studies, reciprocity was a key consideration in which there needed to be mutual value and benefits to the partnership for both myself as a doctoral research student and for the SibYAC. Reciprocity encompasses co-learning (108,117,118), in which the SibYAC and I can learn from each other's expertise and knowledge as a group. I often thought about the personal and group goals shared by each SibYAC member. For example, some SibYAC members wanted to connect to an academic community, and I asked if they were interested and available to co-present presentations at conferences. They wanted to connect with other researchers and asked if my supervisor and graduate supervisory committee members would be interested in attending our monthly SibYAC meetings as a guest. In November 2020, my supervisor (Dr. Jan Willem Gorter) was our first guest followed by the attendance of other members of my graduate supervisory committee members. Prior to our meeting, I provided the agenda, biography of our guest, and Group Meeting Rules (Refer to Appendix B from Chapter 1). Both the guests and SibYAC members provided verbal feedback, in which they valued the conversations to learn from each other. SibYAC members also appreciated in hearing about how siblings have been involved in the research from guests, or how siblings could be potentially incorporated into future research. They also valued the discussions with experts, for example, Dr. Susan facilitated our discussion about data analysis in the BEST SIBS Study. The SibYAC found the discussions therapeutic and were fascinated with learning how their experiences influenced the process of data analysis.

Recommendation 3: Ensure that there is ongoing communication throughout the project.

An additional consideration was to consider different ways to communicate with the SibYAC in a way that they preferred. The primary method of communication was by email to share meeting agendas and summaries, meeting reminders, and share documents for review. I also offered other forms of communication that the SibYAC members preferred, such as a Google Drive folder to review documents and a Facebook group for meeting reminders. I also recognized that there were ongoing initiatives, specifically with the scholarly works in my doctoral studies, and I wanted to provide opportunities for the SibYAC to be engaged in these initiatives. I provided Terms of Reference to outline the three projects with a tentative timeline. The SibYAC could choose to have different roles in these initiatives as outlined in the Involvement Matrix, such as being a listener, co-thinker, advisor, partner, or decision-maker (91). In any of these roles, there was an option for the SibYAC to provide feedback both synchronously and asynchronously, which provides flexibility for the SibYAC to be engaged in research. I wanted to ensure that there was a team, including my graduate supervisory committee and the SibYAC. There may be decisions that were made, such as deadlines for certain projects that needed to be met or limiting the scope of a project. To ensure transparency about the decision-making process, I consistently provided updates to the SibYAC. During meetings, I also had discussions with the SibYAC about how decisions were made and how their feedback was incorporated, or whether there were opportunities for future activities that they may wish to engage in. This transparency helped to ensure that the SibYAC has clear communication about the whole research process, and as a SibYAC member described: “Because I’ve been involved throughout the study and not just after the study ends, I feel truly acknowledged and can see the importance of partnering in research.”

Recommendation 4: Consider multiple ways to acknowledge and appreciate partners.

Partners can be acknowledged and appreciation in different ways that would be meaningful and valuable to them. While compensation is one form of appreciation as described by other patient partners in the literature (110,111), I did not have funding at the beginning of my doctoral studies to provide compensation. We were successful with two grants, the CIHR Patient-Oriented Research Awards – Transition to Leadership Stream – Phase 1 and the CHILD-BRIGHT Graduate Fellowship for Patient-Oriented Research, that provided an allocated budget for compensation. The SibYAC provided their input about how they would like to receive compensation in different formats, such as a combination of an annual honorarium and social activities. Social activities, such as holiday care packages or virtual social gatherings with an Ubereats e-gift card, were important to the SibYAC to enhance our rapport and bond as a team. Conversations about compensation are important, as patient partners may choose to receive their compensation in multiple formats that are valuable to them.

Recommendation 5: Continue to seek opportunities to engage in dialogue and reflections in the field of patient-oriented research.

There are different areas for opportunities to learn about strategies to engage with partnerships. I first acquired about patient-oriented research through the Family Engagement in Research (FER) Certificate of Completion Program hosted by McMaster University Centre for Continuing, CanChild Centre for Childhood Disability Research, and Kids Brain Health Network (119). Since then, I continue to stay connected with current students and graduates from the FER course through a group on Facebook. The group on Facebook allowed me to have ongoing conversations and learn about other patient-oriented initiatives that students and alumni were working on with patient partners. I further sought opportunities to train and learn about patient-oriented research throughout my PhD studies. Supplementary Table 2 describes these training opportunities and the skills that I developed from them. These opportunities can be categorized into: knowledge acquisition, in which I learned about the foundation of patient-oriented research; and experiential learning, in which I applied patient engagement strategies and learned whether these strategies were effective. Through opportunities to acquire and apply knowledge, I was able to continue growing my skills to conduct patient-oriented research in my doctoral studies in partnership with the SibYAC.

Recommendation 6: Engage in reflexivity throughout the partnership.

Researchers and patient partners can engage in reflexivity (105), and have open discussions to build and continue the partnership. Previous researchers, including a team that I was a part of, identified how qualitative research methods can be applied to patient and family engagement (120,121). Reflexivity is a common process that qualitative researchers engage in as a technique to enhance trustworthiness of the data (122). This technique allows researchers to understand and recognize the extent to which their interests, positions, and assumptions influence their research (123). I engaged in reflexivity throughout my partnership with the SibYAC by writing reflexive memos about my thoughts and considerations. Often, these memos were based on topics from the literature about potential barriers and strategies (117,124–126) or potential power imbalances between the SibYAC and myself, which then allowed me to continue to identify ways to create a positive, authentic, and meaningful partnership with the SibYAC. For example, these memos allowed me to understand how to move forward and provide opportunities for the SibYAC to take the lead on initiatives.

Personal next steps

My personal goal is to continue developing my skills to become an independent patient-oriented researcher in the field of childhood disability. I want to continue to build and sustain my partnership with the SibYAC. In my conversations with each SibYAC member, they are motivated to continue our partnership to raise awareness about the important and valuable roles of young adult siblings of individuals with a disability. I plan to expand the diversity of the SibYAC, such as recruiting potential siblings from different cultures and genders. In addition to knowledge translation activities to share the findings from my dissertation, adapt existing resources and co-create components of new resources such as the Sibling Kit, I want to better understand how to engage with siblings, youth, and families at a systems-level and policy-level which I plan to explore during my postdoctoral fellowship.

In closing, the PhD chapter of my journey was only the starting point that fueled my passion for patient-oriented, childhood disability research. I look forward to the next chapter of my journey to explore how to partner with multiple stakeholders in research, healthcare, and policy systems. As an African proverb says, “If you want to go far, go together”.

References

1. Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, et al. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Frontiers in Rehabilitation Sciences. 2021 Oct 5;2:52.
2. Nguyen L, Bootsma J, Rezze B Di, Jack S, Ketelaar M, Gorter JW. Programmes to prepare siblings for future roles to support their brother or sister with a neurodisability: protocol of a scoping review. BMJ Open. 2021;11:53184.
3. Children's Healthcare Canada. About Children's Healthcare Canada [Internet]. [cited 2022 May 7]. Available from: https://www.childrenshealthcarecanada.ca/about
4. Redquest BK, Tint A, Ries H, Goll E, Rossi B, Lunsky Y. Support needs of Canadian adult siblings of brothers and sisters with intellectual/developmental disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2020;17(3):239–46.
5. Cutrona CE. Social support principles for strengthening families: messages from the USA. In: Canavan J, Dolan P, Pinkerton J, editors. Family Support: Direction from Diversity. London: Jessica Kingsley; 2000. p. 103–22.
6. Schaefer C, Coyne JC, Lazarus RS. The health-related functions of social support. Journal of Behavioral Medicine. 1981;4(4):381–406.
7. Giovannetti ER, Wolff JL. Cross-survey differences in national estimates of numbers of caregivers of disabled older adults. Milbank Quarterly. 2010;88(3):310–49.
8. Arnett JJ. Emerging adulthood: A theory of development from the late teens through the twenties. American Psychologist. 2000;55(5):469–80.
9. Burke MM, Arnold CK, Owen AL. Sibling advocacy: perspectives about advocacy from siblings of individuals with intellectual and developmental disabilities. Inclusion. 2015;3(3):162–75.
10. Harland P, Cuskelly M. The responsibilities of adult siblings of adults with dual sensory impairments. International Journal of Disability, Development and Education. 2000;47(3):293–307.
11. Özen A. Effectiveness of siblings-delivered ipad game activities in teaching social interaction skills to children with autism spectrum disorders. Educational Sciences: Theory and Practice. 2015;15(5):1287–303.
12. Fjermestad K, Pat P, Dearozet S, Vatne T, Hafting M, Jegannathan B. Manual-Based Group Intervention for Siblings and Parents of Children with Neurodevelopmental Disorders in Cambodia. Journal of Developmental and Physical Disabilities. 2021;33(5):839–56.
13. The World Bank. The world by income and region [Internet]. 2022 [cited 2022 May 8]. Available from: https://datatopics.worldbank.org/world-development-indicators/the-world-by-income-and-region.html
14. The World Bank. What we do [Internet]. [cited 2022 May 8]. Available from: https://www.worldbank.org/en/what-we-do
15. Lobato DJ, Kao BT. Integrated sibling-parent group intervention to improve sibling knowledge and adjustment to chronic illness and disability. Journal of Pediatric Psychology. 2002;27(8):711–6.
16. Jones EA, Fiani T, Stewart JL, Neil N, McHugh S, Fienup DM. Randomized controlled trial of a sibling support group: Mental health outcomes for siblings of children with autism. Autism. 2020;24(6):1468–81.

17. Meyer DJ, Vadasy PF. Sibshop workshops for siblings of children with special needs. Baltimore: Paul H. Brookes; 1994.
18. Fjermestad KW, Silverman WK, Vatne TM. Group intervention for siblings and parents of children with chronic disorders (SIBS-RCT): study protocol for a randomized controlled trial. Trials. 2020;21(1):1–12.
19. Tsao LL, Odom SL. Sibling-mediated social interaction intervention for young children with autism. Topics in Early Childhood Special Education. 2006;26(2):106–23.
20. Lobato DJ, Kao BT. Brief report: Family-based group intervention for young siblings of children with chronic illness and developmental disability. Journal of Pediatric Psychology. 2005;30(8):678–82.
21. Nguyen L, Jack SM, Di Rezze B, Ketelaar M, Gorter JW. Protocol of the BEST SIBS study: A qualitative case study to investigate the roles and responsibilities of siblings of youth with a neurodisability during health care transition. Journal of Transition Medicine. 2021;3(1):1–11.
22. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. Journal of Applied Research in Intellectual Disabilities. 2018;31(3):423–34.
23. Avieli H, Band-Winterstein T, Araten Bergman T. Sibling relationships over the life course: growing up with a disability. Qualitative Health Research. 2019;29(12):1739–50.
24. Feinberg ME, Solmeyer AR, McHale SM. The third rail of family systems: Sibling relationships, mental and behavioral health, and preventive intervention in childhood and adolescence. Clinical Child and Family Psychology Review. 2012;15(1):43–57.
25. Moyson T, Roeyers H. “The overall quality of my life as a sibling is all right, but of course, it could always be better”. Quality of life of siblings of children with intellectual disability: the siblings’ perspectives. Journal of Intellectual Disability Research. 2012;56(1):87–101.
26. Luijkx J, van der Putten AAJ, Vlaskamp C. “I love my sister, but sometimes I don’t”: A qualitative study into the experiences of siblings of a child with profound intellectual and multiple disabilities†. Journal of Intellectual and Developmental Disability. 2016;41(4):279–88.
27. Kruithof K, IJzerman L, Nieuwenhuijse A, Huisman S, Schippers A, Willems D, et al. Siblings’ and parents’ perspectives on the future care for their family member with profound intellectual and multiple disabilities: A qualitative study. Journal of Intellectual & Developmental Disability. 2021;46(4):351–61.
28. Park S, Ryu W, Yang H. A study on the life experiences of adolescents who grew up with younger siblings with developmental disabilities: Focusing on phenomenological analysis methods. Brain Sciences. 2021;11(6):798.
29. Burke MM, Taylor JL, Urbano R, Hodapp RM. Predictors of future caregiving by adult siblings of individuals with intellectual and developmental disabilities. American Journal on Intellectual and Developmental Disabilities. 2012;117(1):33–47.
30. Lee CE, Burke MM. Caregiving roles of siblings of adults with intellectual and developmental disabilities: a systematic review. Journal of Policy and Practice in Intellectual Disabilities. 2018;15(3):237–46.
31. Saxena M. Variables that can enhance and complicate sibling caregiving of individuals with intellectual and developmental disabilities. Journal of Policy and Practice in Intellectual Disabilities. 2015;12(3):210–9.

32. Burke P. Brothers and sisters of disabled children. London: Jessica Kingsley Publishers; 2004.
33. Burke P. Brothers and sisters of disabled children: The experience of disability by association. British Journal of Social Work. 2010;40(6):1681–99.
34. Hodapp RM, Glidden LM, Kaiser AP. Siblings of persons with disabilities: Toward a research agenda. Mental Retardation. 2005;43(5):334–8.
35. Morris B, Ogden J, Gentle J. Experiences of adult siblings of those with developmental coordination disorder (DCD): A qualitative study. Current Psychology. 2021;1–12.
36. Stoneman Z. Supporting positive sibling relationships during childhood. Mental Retardation and Developmental Disabilities Research Reviews. 2001;7(2):134–42.
37. Leedham AT, Thompson AR, Freeth M. A thematic synthesis of siblings' lived experiences of autism: Distress, responsibilities, compassion and connection. Research in Developmental Disabilities. 2020;97.
38. Watson L, Hanna P, Jones CJ. A systematic review of the experience of being a sibling of a child with an autism spectrum disorder. Clin Child Psychol Psychiatry. 2021;26(3):734–49.
39. Iannuzzi D, Fell L, Luberto C, Goshe BM, Perez G, Park E, et al. Challenges and growth: Lived experience of adolescents and young adults (AYA) with a sibling with ASD. Journal of Autism and Developmental Disorders. 2021;1–8.
40. Perenc L, Pęczkowski R. Cognitive and affective empathy among adolescent siblings of children with a physical disability. Disability and Health Journal. 2018;11(1):43–8.
41. Rossetti Z, Lee C eun, Burke M, Hall S. Perspectives about adult sibling relationships: A dyadic analysis of siblings with and without intellectual and developmental disabilities. Research in Developmental Disabilities. 2020;96:103538.
42. Begum G, Blacher J. The siblings relationship of adolescents with and without intellectual disabilities. Research in Developmental Disabilities. 2011;32(5):1580–8.
43. Flaton RA. "Who would I be without Danny?" Phenomenological case study of an adult sibling. Mental Retardation. 2006;44(2):135–44.
44. Hodapp RM, Sanderson KA, Meskis SA, Casale EG. Adult siblings of persons with intellectual disabilities: Past, present, and future. International Review of Research in Developmental Disabilities. 2017;53:163–202.
45. Marks SU, Matson A, Barraza L. The impact of siblings with disabilities on their brothers and sisters pursuing a career in special education. Research and Practice for Persons with Severe Disabilities. 2005;30(4):205–18.
46. Graham ID, Logan J, Harrison MB, Straus SE, Tetroe J, Caswell W, et al. Lost in Knowledge Translation: Time for a Map? Journal of continuing education in the health professions. 2006;26(1):13–24.
47. CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth wIth Brain-based disabilitieS (BEST SIBS) Study [Internet]. 2021 [cited 2022 Feb 4]. Available from: https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study
48. Children's Healthcare Canada. Health Hub in Transition [Internet]. 2021 [cited 2021 Mar 31]. Available from: https://www.transitionhub.ca/
49. Cross A, Rosenbaum P, Grahovac D, Brocklehurst J, Kay D, Baptiste S, et al. A web-based knowledge translation resource for families and service providers (the "F-words"

in childhood disability knowledge hub): Developmental and pilot evaluation study. JMIR Rehabil Assist Technol. 2018;5(2):e10439.

50. Freeman M, Stewart D, Shimmell L, Missiuna C, Burke-Gaffney J, Jaffer S, et al. Development and evaluation of The KIT: Keeping It Together^{TM}forYouth (the 'Youth KIT') to assist youth with disabilities in managing information. Child: Care, Health and Development. 2015;41(2):222–9.
51. Stewart D, Law M, Burke-Gaffney J, Missiuna C, Rosenbaum P, King G, et al. Keeping it together^{TM}: An information KIT for parents of children and youth with special needs. Child: Care, Health and Development. 2006;32(4):493–500.
52. Holland Bloorview Kids Rehabilitation Hospital. SibKits [Internet]. [cited 2021 May 29]. Available from: https://hollandbloorview.ca/services/family-workshops-resources/family-resource-centre/online-family-resources-centre/sibkits
53. Whiteman SD, McHale SM, Soli A. Theoretical perspectives on sibling relationships. Journal of Family Theory & Review. 2011;3(2):124–39.
54. Siddiqui A, Ross H. Mediation as a method of parent intervention in children's disputes. Journal of Family Psychology. 2004;18(1):147–59.
55. Perlman M, Ross HS. The benefits of parent intervention in children's disputes: An examination of concurrent changes in children's fighting styles. Child Development. 1997;68(4):690.
56. Davies MD, Beamish W. Transitions from school for young adults with intellectual disability: Parental perspectives on "life as an adjustment." Journal of Intellectual and Developmental Disability. 2009;34(3):248–57.
57. Stewart DA, Law MC, Rosenbaum P, Willms DG. A qualitative study of the transition to adulthood for youth with physical disabilities. Physical and Occupational Therapy in Pediatrics. 2001;21(4):3–21.
58. Kingsnorth S, Rudzik AEF, King G, McPherson AC. Residential immersive life skills programs for youth with disabilities: A case study of youth developmental trajectories of personal growth and caregiver perspectives. BMC Pediatrics. 2019;19(1):413.
59. Bronfenbrenner U. Six theories of child development: Revised formulations and current issues. Vasta R, editor. London: Jessica Kingsley Publishers; 1992. 187–249 p.
60. Heller T, Kramer J. Involvement of adult siblings of persons with developmental disabilities in future planning. Intellectual and Developmental Disabilities. 2009;47(3):208–19.
61. Davys D, Mitchell D, Haigh C. Futures planning - adult sibling perspectives. British Journal of Learning Disabilities. 2015;43(3):219–26.
62. Leane M. "I don't care anymore if she wants to cry through the whole conversation, because it needs to be addressed": Adult siblings' experiences of the dynamics of future care planning for brothers and sisters with a developmental disability. Journal of Applied Research in Intellectual Disabilities. 2020;33(5):950–61.
63. Noonan H, O'Donoghue I, Wilson C. Engaging with and navigating limbo: Lived experiences of siblings of adults with autism spectrum disorders. Journal of Applied Research in Intellectual Disabilities. 2018;31(6):1144–53.
64. Hagiwara M, Palmer SB, Hancock CL, Shogren KA. Sibling roles in family–school partnerships for students with disabilities during transition planning. Career Development and Transition for Exceptional Individuals. 2019;42(3):194–200.

65. Arriagada P, Statistics Canada. Insights on Canadian Society: The experiences and needs of older caregivers in Canada [Internet]. 2020 [cited 2022 May 8]. Available from: https://www150.statcan.gc.ca/n1/pub/75-006-x/2020001/article/00007-eng.htm
66. Torjman S. Policies in support of caregivers [Internet]. 2015 [cited 2022 May 8]. Available from: https://munkschool.utoronto.ca/mowatcentre/wp-content/uploads/publications/RCSA_policies_in_support_of_caregivers.pdf
67. Carers Canada. About Us [Internet]. [cited 2022 May 8]. Available from: https://www.carerscanada.ca/about-us/
68. Ontario Caregiver Coalition. About Us [Internet]. [cited 2022 May 8]. Available from: https://www.ontariocaregivercoalition.ca/
69. Young Carers Program. Mission and goals [Internet]. [cited 2022 May 8]. Available from: https://www.ycptoronto.com/who-we-are.html
70. National Health Services. Being a young carer: Your rights [Internet]. 2021 [cited 2022 May 8]. Available from: https://www.nhs.uk/conditions/social-care-and-support-guide/support-and-benefits-for-carers/being-a-young-carer-your-rights/
71. Young Carers Program. What is a young carer? [Internet]. [cited 2022 May 8]. Available from: https://www.ycptoronto.com/young-carer.html
72. Australian Government. National Carer Strategy [Internet]. 2011. Available from: https://www.carerstas.org/wp-content/uploads/2017/06/2011-National-Carer-Strategy.pdf
73. Canadian Centre for Caregiving Excellence. Siblings Canada [Internet]. 2022 [cited 2022 May 8]. Available from: https://canadiancaregiving.org/siblingscanada/
74. Yin RK. Designing case studies. In: Case Study Research and Applications: Design and Methods. 6th ed. Thousand Oaks, California: SAGE; 2018. p. 25–80.
75. Yin RK. The Case Study Crisis: Some Answers. Administrative Science Quarterly. 1981;26(1):58.
76. Yin RK. Enhancing the quality of case studies in health services research. Health Services Research [Internet]. 1999 [cited 2022 Jul 9];34(5 Pt 2):1209. Available from: /pmc/articles/PMC1089060/?report=abstract
77. Meleis AI, Sawyer LM, Im EO, Messias DK, Schumacher K. Experiencing transitions: an emerging middle-range theory. Advances in Nursing Science. 2000;23(1):12–28.
78. Baxter P, Jack S. Qualitative case study methodology: Study design and implementation for novice researchers. The Qualitative Report. 2008;13(2):544–59.
79. Yin RK. Designing case studies: Identifying your case(s) and establishing the logic of your case study. In: Case Study Research and Applications: Design and Methods. 6th ed. Thousand Oaks, California: Sage Publications Inc.; 2018. p. 25–78.
80. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. Canadian Journal of Community Mental Health. 2014;33(1):45–61.
81. Palisano RJ, Di Rezze B, Stewart D, Rosenbaum PL, Hlyva O, Freeman M, et al. Life course health development of individuals with neurodevelopmental conditions. Developmental Medicine & Child Neurology. 2017;59(5):470–6.
82. Yin RK. Analyzing case study evidence. In: Case study research and applications: Design and methods. 6th ed. Thousand Oaks: Sage Publications, Inc.; 2018. p. 165–218.

83. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology [Internet]. 2006 [cited 2019 Oct 9];3(2):77–101. Available from: http://www.tandfonline.com/doi/abs/10.1191/1478088706qp063oa
84. Lincoln Y, Guba EG. Naturalistic inquiry. Beverly Hills: SAGE; 1985.
85. Guba EG, Lincoln YS. Fourth generation evaluation. Newbury Park: SAGE; 1989.
86. Barber JP, Walczak KK. Conscience and critic: Peer debriefing strategies in grounded theory research. Ann Arbor; 2009.
87. Freysteinson WM, Lewis C, Sisk A, Wuest L, Deutsch AS, Cesario SK. Investigator reflections: A final debriefing following emotionally sensitive mirror research. Holistic Nursing Practice. 2013;27(3):177–84.
88. Richards KAR, Hemphill MA. A practical guide to collaborative qualitative data analysis. Journal of Teaching in Physical Education. 2018;37(2):225–31.
89. Spall S, Austin SF. Peer debriefing in qualitative research: Emerging operational models.
90. Shenton AK. Strategies for ensuring trustworthiness in qualitative research projects. Education for Information. 2004;22(2):63–75.
91. Smits DW, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. Research Involvement and Engagement. 2020;6(1):30.
92. Richards DP, Birnie KA, Eubanks K, Lane T, Linkiewich D, Singer L, et al. Guidance on authorship with and acknowledgement of patient partners in patient-oriented research. Research Involvement and Engagement. 2020;6(1):38.
93. Robson C. Real world research: A resource for social scientists and practitioner-researchers. 2nd ed. Oxford: Wiley-Blackwell; 2002. p. 22.
94. McGhee G, Marland GR, Atkinson J. Grounded theory research: Literature reviewing and reflexivity. Journal of Advanced Nursing. 2007;60(3):334–42.
95. Backman K, Kyngas HA. Challenges of the grounded theory approach to a novice researcher. Nursing and Health Sciences. 1999;1(3):147–53.
96. Carcary M. The research audit trail: Enhancing trustworthiness in qualitative inquiry. Electronic Journal of Business Research Methods. 2009;7(1):11–24.
97. Rodgers BL, Cowles K V. The qualitative research audit trail: A complex collection of documentation. Research in Nursing & Health. 1993;16(3):219–26.
98. Salvatori P, Tremblay M, Tryssenaar J. Living and aging with a developmental disability: Perspectives of individuals, family members and service providers. Journal on developmental disabilities. 2003;10(1):1–20.
99. Font SA, Kim HW. Sibling separation and placement instability for children in foster care. Child Maltreatment. 2021;1–13.
100. Linares LO, Li M, Shrout PE, Brody GH, Pettit GS. Placement shift, sibling relationship quality, and child outcomes in foster care: A controlled study. Journal of Family Psychology. 2007;21(4):736–43.
101. Abrams JA, Tabaac A, Jung S, Else-Quest NM. Considerations for employing intersectionality in qualitative health research. Social Science & Medicine. 2020;258:113138.
102. Kimberle. C. Demarginalizing the intersection of race and sex: A Black feminist critique of antidiscrimination doctrine, feminist theory, and antiracist politics. In: Katharine T.B & Rosanne. K, editor. Feminist Legal Theory: Readings in Law and Gender. Routledge; 1989. p. 57–80.

103. Else-Quest NM, Hyde JS. Intersectionality in quantitative psychological research: I. Theoretical and epistemological issues. Psychology of Women Quarterly. 2016;40(2):155–70.
104. Lee CE, Hagiwara M, Black H. A scoping review of cross-cultural experiences of siblings of individuals with intellectual and developmental disabilities in the United States. Research in Developmental Disabilities. 2021;112(2021):103916.
105. Staley K. 'Is it worth doing?' Measuring the impact of patient and public involvement in research. Research Involvement and Engagement. 2015;1(1):1–10.
106. Dawson S, Ruddock A, Parmar V, Morris R, Cheraghi-Sohi S, Giles S, et al. Patient and public involvement in doctoral research: Reflections and experiences of the PPI contributors and researcher. Research Involvement and Engagement. 2020;6(1):1–13.
107. Coupe N, Mathieson A. Patient and public involvement in doctoral research: Impact, resources and recommendations. Health Expectations. 2020;23(1):125–36.
108. Canadian Institutes of Health Research. Strategy for Patient Oriented Research. Patient engagement framework [Internet]. 2014. Available from: http://www.cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf
109. Sibling Youth Advisory Council. Publications and Presentations. 2022.
110. Richards DP, Cobey KD, Proulx L, Dawson S, de Wit M, Toupin-April K. Identifying potential barriers and solutions to patient partner compensation (payment) in research. Research Involvement and Engagement. 2022;8(1):7.
111. Richards DP, Jordan I, Strain K, Press Z. Patient partner compensation in research and health care: the patient perspective on why and how. Patient Experience Journal. 2018;5(3):6–12.
112. CHILD-BRIGHT Network. Guidelines for Patient Partner Compensation [Internet]. 2020 [cited 2022 Apr 6]. Available from: https://www.child-bright.ca/compensation-guidelines
113. Canadian Institutes of Health Research. Considerations when paying patient partners in research [Internet]. 2021 [cited 2022 Feb 8]. Available from: https://cihr-irsc.gc.ca/e/51466.html
114. Cassidy CE, Shin HD, Ramage E, Conway A, Mrklas K, Laur C, et al. Trainee-led research using an integrated knowledge translation or other research partnership approaches: a scoping reviews. Health Research Policy and Systems [Internet]. 2021 Dec 1 [cited 2022 Jul 9];19(1):1–50. Available from: https://link.springer.com/articles/10.1186/s12961-021-00784-0
115. Abelson J, Li K, Wilson G, Shields K, Schneider C, Boesveld S. Supporting quality public and patient engagement in health system organizations: development and usability testing of the Public and Patient Engagement Evaluation Tool. Health Expectations. 2016;19(4):817–27.
116. de Wit MPT, Elberse JE, Broerse JEW, Abma TA. Do not forget the professional - the value of the FIRST model for guiding the structural involvement of patients in rheumatology research. Health Expectations. 2015;18(4):489–503.
117. Black A, Strain K, Wallsworth C, Charlton SG, Chang W, McNamee K, et al. What constitutes meaningful engagement for patients and families as partners on research teams? Journal of Health Services Research and Policy. 2018;23(3):158–67.
118. Salerno J, Coleman KJ, Jones F, Peters ES. The ethical challenges and opportunities of implementing engagement strategies in health research. Annals of Epidemiology. 2021;59:37–43.

119. CanChild Centre for Childhood Disability Research, Kids Brain Health Network, McMaster University. Family Engagement in Research Course [Internet]. 2021 [cited 2021 Nov 7]. Available from: https://www.canchild.ca/en/research-in-practice/family-engagement-in-research-course
120. Phoenix M, Nguyen T, Gentles SJ, VanderKaay S, Cross A, Nguyen L. Using qualitative research perspectives to inform patient engagement in research. Research Involvement and Engagement. 2018;4(1):20.
121. Rolfe DE, Ramsden VR, Banner D, Graham ID. Using qualitative health research methods to improve patient and public involvement and engagement in research. Research Involvement and Engagement. 2018;4(1):1–8.
122. Finlay L. "Outing" the researcher: The provenance, process, and practice of reflexivity. Qualitative Health Research. 2002;12(4):531–45.
123. Charmaz K. Constructing grounded theory: A practical guide through qualitative analysis. London: Sage Publications; 2006.
124. Harrison JD, Anderson WG, Fagan M, Robinson E, Schnipper J, Symczak G, et al. Patient and Family Advisory Councils (PFACs): Identifying challenges and solutions to support engagement in research. Patient. 2018;11(4):413–23.
125. Rouleau G, Bélisle-Pipon JC, Birko S, Karazivan P, Fernandez N, Bilodeau K, et al. Early career researchers' perspectives and roles in patient-oriented research. Research Involvement and Engagement. 2018;4(1):1–15.
126. Hahn DL, Hoffmann AE, Felzien M, LeMaster JW, Xu J, Fagnan LJ. Tokenism in patient engagement. Family Practice. 2017;34(3):290–5.

References for Figure 1

1. Nguyen L, Davis H, Bellefeuille S. CHILD-BRIGHT Virtual Symposium Part 2 [Internet]. 2020 [cited 2022 Mar 9]. Available from: https://www.child-bright.ca/events/2020/4/29/symposium-pt2
2. Backlin G, Bellefeuille S, Davis H, Lolonde C, Nguyen L, Tesolin J. Learning from youth and young adults: Engagement strategies for patient-oriented research projects, committees, and councils [Internet]. 2020 [cited 2022 Mar 9]. Available from: https://www.child-bright.ca/events/2020/9/9/youth-engagement-strategies
3. CHILD-BRIGHT National Youth Advisory Panel and CanChild Sibling Youth Advisory Council. 10 Tips for Engaging Young People in Research [Internet]. 2020 [cited 2022 Mar 9]. Available from: https://www.child-bright.ca/nyap-sibyac-tips-for-researchers
4. Sibling Youth Advisory Council. SibYAC Activities [Internet]. 2022 [cited 2022 Mar 9]. Available from: https://www.canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study/sibyac-activities
5. Bellefeuille S, Davis H, Havens J, Nguyen L. CanChild Luke's Legacy Family Research Rounds: The Art of Sibling Science [Internet]. 2021 [cited 2022 Mar 2]. Available from: https://vimeo.com/543787118
6. Nguyen L, Davis H, Bellefeuille S, Havens J, Jack SM, Di Rezze B, et al. Canadian resources for siblings of youth with chronic health conditions to inform and support with healthcare management: A qualitative document analysis. Front Rehabil Sci. 2021;2:52.

Supplementary Table 1. Examples of how the Sibling Youth Advisory Council and I demonstrated key values of our partnership.

Value	**Examples displayed by both the SibYAC and myself**	**Examples displayed by myself as the facilitator**
Respect	Verbal acknowledgement of each member's ideas; we further provided our thoughts or alternative ideas.	
Understanding	Recognition that everyone had different schedules and may not be able to join team meetings. When individuals were able to attend meetings, we valued their contributions and welcomed each SibYAC member to join team meetings when they can.	I encouraged every SibYAC member to let me know if they would like an individual check-in meeting. All SibYAC members were welcome to provide their input asynchronously for initiatives.
Appreciation	Each SibYAC member stated that they appreciated the opportunity to be engaged in different initiatives.	I had discussions about ways that the SibYAC would like to have tokens of appreciation (e.g., annual holiday care packages of customized mugs and/or cookies, virtual holiday gatherings, Ubereats e-gift cards). My emails also included a message to thank the SibYAC for their ongoing support and partnership in my doctoral studies.

Supplementary Table 2. Description of training opportunities.

Training Opportunity	**Organization**	**Date**	**Description**	**Development of Skills**
Knowledge Acquisition				
Family Engagement in Research Certificate of Completion Program (1)	Centre for Continuing Education, CanChild Centre for Childhood Disability Research, and Kids Brain Health Network	Jan 2019 – Mar 2019	A 10-week course delivered online for researchers and families interested in child neurodevelopmental research.	• I developed a foundational understanding about patient-oriented research, including its importance, strategies of how to engage with families, barriers and facilitators to engagement, ethics, and tools and resources to support and evaluate engagement.
Webinars	CHILD-BRIGHT Network	Started since 2018 Sept 2021 – Mar 2022	Learning Series (2) A series of webinars hosted by multiple research teams to share experiences of engaging families in research, including the barriers and facilitators. Patient-Oriented Research Discussions (3) These series of online discussion were hosted by award recipients of the Graduate Fellowship in Patient-Oriented Research from the CHILD-BRIGHT Network along with parent partners. Discussion topics include experiences of	• I acquired further knowledge about strategies to engage with families in research based on the experiences from different research projects. • I had the opportunity to co-facilitate these patient-oriented research discussions, in which I shared about my experiences as a trainee conducting patient-oriented research and learned from other researchers and family partners about their engagement in research. These discussion topics were also relevant to

			graduate students and new researchers, practical tools and training resources that incorporate patient-oriented methodologies, integration of the patient perspectives in data analysis, and topical issues in patient-oriented research using simulated scenarios.	identify current challenges that we were facing in our projects, such as how to incorporate the perspectives of patient partners in data analysis, and potential strategies to these challenges.
Workshops	School of Rehabilitation Science at McMaster University	April 14, 2021 Monthly attendance since 2015	*Skills Series* Monthly skills series are held for graduate students in the School of Rehabilitation Science to learn and develop research skills. One of the skills series was facilitated by Dr. Michelle Phoenix, an Assistant Professor in the Speech-Language Pathology Program in the School of Rehabilitation Science at McMaster University about community-engaged research. *Qualitative Query* Qualitative query sessions are held monthly to discuss qualitative methodologies.	• At this skills series facilitated by Dr. Michelle Phoenix, I learned about the strategies that she used to engage with community partners, as well as important considerations for how to develop an authentic and meaningful relationship between researchers and community partners. • At one of the Qualitative Query sessions prior to starting my PhD, I had a conversation with my peers about how qualitative methods can be applied to patient-oriented research. Based on this discussion, the team and I wrote a research paper (4), and I continued to apply qualitative methods in my

				partnership with the SibYAC such as being reflexive and having peer debriefings during data analysis.
Mentorship	With Dr. Marjolijn Ketelaar, an Associate Professor at the Center of Excellence for Rehabilitation Medicine Utrecht, the research institute of the UMC Utrecht Brain Center, and De Hoogstraat Rehabilitation	Sept 2017 – present	Dr. Marjolijn Ketelaar has been a member of my graduate supervisory committee. She is also the Co-Principal Investigator of the PiP-PERRIN Study Group with extensive experience in engagement with youth ambassadors in research studies (5).	• In April 2020, I had planned to embark on an exchange to the Netherlands to further learn about how to engage with youth patients and families in research. Due to the COVID-19, this exchange was placed on hold but I had the opportunity to continue receiving mentorship from Dr. Marjolijn Ketelaar virtually. We had conversations, in which I learned about different ways that she engaged with youth ambassadors with cerebral palsy in research and family partners in other research projects, and she learned about how I was partnering with the SibYAC. One strategy that I learned is about the importance of having fun, and she described how youth ambassadors wanted to attend social activities, such

				as cooking workshops, that allowed them to bond with each other in a non-research context. She shared about the importance of developing a relationship with partners outside of their role as an ‘expert’ by their lived experiences, but also about getting to know about their family, hobbies, and dreams which then allows for the development of the whole team. Based on her experiences, we came up with the idea to have a virtual holiday get-together in which I mailed holiday care packages to each SibYAC member, and we met virtually to open the packages together and had an informal group conversation. This event further strengthened our partnership.
Conversations	Varied	Sept 2017 – present	Conversations with experts in the field of patient-oriented research.	• I sought opportunities to have conversations with experts in the field of patient-oriented research. For example, I consulted with Dr. Keiko Shikako who is the co-lead of the

				CHILD-BRIGHT Knowledge Translation Committee to identify appropriate tools to evaluate patient engagement. • I also reached out to the research team who developed the Public and Patient Engagement Evaluation Tool (PPEET, 6) to ask about how this tool can be applied to my partnership with the SibYAC. The team described that the tool can be completed within 15 minutes. I learned from the team about the importance of identifying a clear purpose for the tool. I realized that for the purpose of evaluating my partnership with the SibYAC, the PPEET could be used to identify strengths and areas of improvement, and have subsequent discussions as feedback for the partnership moving forward.

Experiential Learning				
Graduate Research Assistant	Patient and Family Advisory Council (PFAC)	Sept 2017 – Dec 2021	Provided support with activities with one of the projects in the CHILD-BRIGHT Network: READiness in Youth fOR transition Out of pediatric Care Brain-Based Disabilities (READYorNot™ BBD) Project	• I learned from the research team and provided support with facilitating the PFAC, including meetings (e.g., inviting guest speakers to open the lines of communication between members of the randomized controlled trial team and the PFAC) and time to have conversations on topics that PFAC members found to be important. • I learned how to develop rapport with the PFAC through individual check-in meetings that are held annually using the ‘Start, Stop, Continue’ activity to discuss what activities should we start, stop, or continue doing, as well as the Involvement Matrix (7). • I provided support with creating documents to onboarding new members to the PFAC, including a Terms of Reference and an initial meeting to orient members to the project. • I learned how to ensure transparency, such as

				closing the loop with sharing how feedback from the PFAC was incorporated into the project. • I co-created knowledge translation products with the PFAC, such as a stakeholder engagement video with testimonials about engagement in the project, as well as a research video series (8) • Ensuring reciprocity, in which there were opportunities of value to PFAC members, such as attending conferences or being engaged in meaning initiatives such as policy work.
Co-presentations and networking at conferences	1. CHILD-BRIGHT Virtual Symposium (9,10) 2. American Academy of Cerebral Palsy and Developmental Medicine (11–14) 3. European Academy of Childhood Disability (15) 4. Australasian Academy of Cerebral Palsy and Developmental Medicine (AusACPDM) and International Alliance of	Sept 2017 – Mar 2022	Co-presentations and networking opportunities at local, national, and international conferences. These co-presentations were conducted in partnership with members of the PFAC or SibYAC.	• When I planned for these co-presentations, I learned about the importance of being flexible to partner with youth patient, sibling, and family partners. I recognize that everyone has different schedules and commitments, and some partners might not be available to present synchronously with me. An approach that I used was to

	Academies of Childhood Disability (IAACD) (16) 5. Canadian Transitions Pop-Up Event (17–19) 6. Kids Brain Health Network Conference (20) 7. CP-Net Science and Family Day (21)			record the presentations, and partners could join if they were available for the question-and-answer period. It was also important to consider whether the registration fee for partners could be covered. Before I received funding through the CIHR Fellowship: Patient-Oriented Research Award – Transition to Leadership Stream (Phase 1) and the Graduate Fellowship in Patient-Oriented Research from the CHILD-BRIGHT Network to provide compensation to SibYAC members, I advocated to conferences to ask for a waiver or reduced cost in registration fee for partners. • I also further connected and learned from other researchers and trainees engaged in patient-oriented research.
Member of the Training Committee	CHILD-BRIGHT Network	Sept 2019 – present	The Training Committee is responsible for establishing, planning, and hosting training initiatives to promote patient-oriented research in the	• I have had the opportunity to learn about training initiatives in patient-oriented research at a network level. I also share

			CHILD-BRIGHT Network. These initiatives include workshops, webinars, and funding opportunities for youth and parent partners, researchers, and other stakeholders in the network.	my experiences as a doctoral trainee about activities and initiatives that would be helpful to further develop patient-oriented research skills for other trainees similar to me.

References for Supplementary Table 2

1. CanChild Centre for Childhood Disability Research, Kids Brain Health Network, McMaster University. Family Engagement in Research Course [Internet]. 2021 [cited 2021 Nov 7]. Available from: https://www.canchild.ca/en/research-in-practice/family-engagement-in-research-course
2. CHILD-BRIGHT Network. CHILD-BRIGHT Events [Internet]. 2019 [cited 2022 Mar 24]. Available from: https://www.child-bright.ca/events
3. CHILD-BRIGHT Network. Patient-oriented research discussions [Internet]. 2019 [cited 2022 Mar 24]. Available from: https://www.child-bright.ca/new-blog/2021/8/24/pods
4. Phoenix M, Nguyen T, Gentles SJ, VanderKaay S, Cross A, Nguyen L. Using qualitative research perspectives to inform patient engagement in research. Res Involv Engagem. 2018;4(1):20.
5. Wintels SC, Researcher J, Smits D. How do adolescents with cerebral palsy participate ? Learning from their personal experiences. 2018;(April):1–11.
6. Abelson J, Li K, Wilson G, Shields K, Schneider C, Boesveld S. Supporting quality public and patient engagement in health system organizations: development and usability testing of the Public and Patient Engagement Evaluation Tool. Heal Expect. 2016;19(4):817–27.
7. Smits D-W, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the Involvement Matrix. Res Involv Engagem. 2020;6(1):30.
8. CHILD-BRIGHT Network. READYorNot(TM) Brain-Based Disabilities Project [Internet]. 2019 [cited 2020 Nov 15]. Available from: https://www.child-bright.ca
9. Nguyen L, Pozniak K, Chambers K. CHILD-BRIGHT Virtual Symposium Part 1 [Internet]. 2020 [cited 2020 Jul 6]. Available from: https://www.child-bright.ca/events/2020/5/15/symposium-pt1
10. Nguyen L, Davis H, Bellefeuille S. CHILD-BRIGHT Virtual Symposium Part 2 [Internet]. 2020 [cited 2022 Mar 9]. Available from: https://www.child-bright.ca/events/2020/4/29/symposium-pt2
11. Nguyen L, Dawe-McCord C, Galuppi B, Mahlberg N, Strohm S, Thomson D, Via-Dufresne Ley A, Kovacs A, Rozenblum R, Marelli A, Gorter JW on behalf of the READYorNot™ Brain-Based Disabilities Project. Ready or not, let's go: Partnering with youth and families to conduct the READYorNot^{TM} Brain-Based Disabilities trial during a pandemic [Demonstration poster]. American Academy for Cerebral Palsy and Developmental Medicine; 2021.
12. Gorter JW, Thomson D, Amaria K, Rozenblum R, Strohm S, Nguyen L, Galuppi B, & Marelli A for the READYorNot™ Brain-Based Disabilities Project. Meaningful involvement of patient, family and healthcare stakeholders in the READYorNot(TM) project [Demonstration poster]. Cincinatti, Ohio: American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) Conference; 2018.
13. Nguyen L, Davis H, Ketelaar M, Di Rezze B, Jack S, Gorter J. Revealing the potential of sibling engagement in research [Demonstration poster]. American Academy for Cerebral Palsy and Developmental Medicine; 2020.
14. Nguyen L, Nguyen T, Shikako K, Gorter JW. Unlocking the potential of youth engagement in childhood disability research [Lunch and learn seminar]. American Academy for Cerebral Palsy and Developmental Medicine; 2020.
15. Nguyen L, Davis H, Bellefeuille S, Ketelaar M, Di Rezze B, Jack S, et al. Growing and

learning together: The process and outcomes of partnering with siblings to design a doctoral patient-oriented research study [Oral presentation]. European Academy of Childhood Disability Europe 2021 Thematic Day: Care, Research and Innovation, the new Landscape of User-professional Partnership; 2021.

16. Nguyen L, van Oort B, van der Meulen E, Davis HD, Bellefeuille S, Matthews J, et al. Better research together: Conversations with researchers and young people [Virtual symposium]. Australasia Academy of Cerebral Palsy and Developmental Medicine and the International Alliance of Academies of Childhood Disabilities Conference 'Better Together' (AusACPDM/IAACD 2022); 2022.
17. Nguyen L, Pozniak K, Chambers K, Galuppi B, Mahlberg N, Via-Dufresne Ley A, Doucet S, Marelli A, Kovacs A, Amaria K, Rozenblum R, Gorter JW on behalf of the READYorNot™ Brain-Based Disabilities Project. Go far, go together: A journey of collaboration with youth and families for the READYorNot(TM) Brain-Based Disabilities Trial [Oral presentation]. Canadian Transitions Pop-Up Event; 2021.
18. Nguyen L, Ketelaar M, Di Rezze B, Jack S & Gorter JW in partnership with the SYAC. The BEST SIBS Study: A qualitative case study to understand the experiences of siblings of youth with a brain-based disability during health care transition [poster display]. Canadian Transitions Pop-Up Event; 2021.
19. Nguyen L, Davis H, Bellefeuille S, Ketelaar M, Di Rezze B, Jack S, et al. Growing together: The process and initial outcomes of partnering with siblings in a doctoral patient-oriented research study on health care transition. Canadian Transitions Pop-Up Event; 2021.
20. Nguyen L, Davis H, Bellefeuille S, Ketelaar M, Di Rezze B, Jack S, et al. Growing together: The process and initial outcomes of partnering with siblings in a doctoral patient-oriented research study on transition [Poster presentation]. Kids Brain Health Network Conference; 2020.
21. Nguyen L, Davis H, Ketelaar M, Di Rezze B, Jack S, Gorter JW. Revealing the potential of sibling engagement in research [Poster presentation]. CP-Net Science and Family Day; 2020.