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EXPERIENCES OF YOUNG ADULTS WITH PEDIATRIC LIFE
THREATENING CONDITIONS TRANSITIONING FROM PEDIATRIC TO
ADULT SERVICES:
A CASE STUDY

By

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ABSTRACT

Introduction

A new group of medically fragile young adults with conditions such as Duchenne's Muscular Dystrophy (DMD) is graduating from pediatric palliative care programs with limited expectations to live beyond early adulthood, and there are very limited comparable adult health and social services to support their complex needs. When these young adults leave supportive pediatric services, they are poorly served by adult services that are not prepared to accommodate to their needs. Accessing this population is difficult because of the complexity of the individuals' conditions, their limited mobility to travel and commit to specific interview times, and divergent communication abilities due to weakness, that lead to the use of computer mediated voice recognition, typing or speech.

Purpose

This case study is among the first research articles to describe the experiences of young adults with pediatric life threatening conditions (pedLTC) who have transitioned from pediatric to adult services. Further, to engage this population of medically fragile participants in the research process, an innovative online data collection method, a bulletin board focus group (BBFG), was utilized.

Methods

A descriptive single case study design with embedded units was selected to examine the complexity of the transition process for young adults with pedLTC.

Face-to-face interviews or bulletin board focus groups were conducted with 10 young adults with pedLTC and 4 expert consultants. Reviews of six documents were also conducted. Conventional and directed qualitative content analysis was conducted on the interview transcripts and documents; and the existing literature and clinical expert opinions were used to triangulate, develop and confirm final interpretations.

Results

After nearly a decade of effort toward implementing programs, protocols and policies to improve transition from pediatric to adult care, very limited progress in new services has been made. Current resources in the adult health, education, and social service systems are inadequate to meet the needs of young adults with pedLTC. Some of the participants in this study demonstrated a striking capacity to navigate through system barriers, whereas others “got stuck” and were delayed and thwarted from pursuing their ambitions. Bulletin board focus groups facilitated an innovative means to reach medically fragile populations and offered creative and richly contextual data.

Conclusions

System resources are imperative to provide the necessary resources such as counselling and mental health services, support for educational and vocational opportunities, development of community programs, and socioeconomic improvement. Connectedness across levels of practice and policy, and education, health and social systems is important to create synergistic effects of intervention.

Young adult hospice programs can provide the resilience lifelines required to counter the risks associated with declining health, and decreasing support from health, social and educational services to support young adults with pedLTC to pursue their ambitions.

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I am grateful to have had the privilege to embark on this PhD journey as a “mature” student and late-ish in my career. The path to the finish had unexpected twists and turns, including a move across the country and resettling my family; the declining health and death of my father; many “near” end of life experiences with my mother; all three of my children graduating from high school and moving away to university; and a skiing accident that immobilized me for months, and fortuitously forced me to stay at home and write this dissertation.

There are many people to thank for their support, encouragement and mentorship. Nearly two decades ago, my friend and mentor, Dr. Betty Davies, planted the PhD seed, and her encouragement of my abilities eventually germinated, and ultimately took root. I have been privileged with a supervisory committee, Drs. Gina Browne, Susan Jack, Lehana Thabane, and Hal Siden, who followed my musings down several detour routes (and across the country!) before settling on this dissertation topic. Gina’s creative approach to health care problems, her enthusiastic and substantive expansion of developing ideas, and kind encouragement have sustained me through this journey. Susan’s amazing support in qualitative methods and analysis, and navigating successfully through the academic system was invaluable. Lehana’s incisive and helpful direction was always appreciated, and Hal graciously supported my research endeavours with Canuck Place Children’s Hospice graduates and offered significant clinical

insights. Thanks as well to Susan Poitras, clinical counsellor, who knows and is deeply committed to all the young adults who have graduated from Canuck Place.

Family and friends have resolutely stood by me through the sagas of graduate school as a mother, wife, daughter and friend. This work could not have been accomplished without the support of my husband Russ, who has been steadfast, incredibly generous, and unfailingly supportive from the inception of the idea to embark on a PhD journey, to believing in me when the mountain felt “too big to climb”, and being good company, my first reader, and a listening ear to the very end of this endeavour. I have enormous appreciation for my three amazing children: Alex, Liam, and Amelia who not only coped, but thrived in the midst of my distraction and commitment to this project. Amelia lived at home for the entire span of my PhD studies and patiently bore with my late arrivals as I finished up “just one more thing”. I am immensely proud of who all of you are, and your amazing gifts. And many thanks to my kind and supportive friends, who were graciously curious about my research, listened to my laments, made me laugh, and cheered me on.

Finally, and most importantly, I want to thank all of the young adults who shared their stories and experiences with me. I am sincerely honoured, and admire your strength, perseverance and wisdom. Your thoughts and comments were a constant marvel and truly inspirational. These pages are dedicated to you with the intent that by telling your stories, transformative system changes will support your courageous and creative life investments.

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Preface

This sandwich thesis includes three manuscripts intended for publication in a peer-reviewed journal. Chapter 5 (*Life Investments*) represents a manuscript prepared for future submission. At the time of writing, Chapter 3, (*Innovations in Research with Medically Fragile Populations*), has been submitted for publication, awaiting an editorial review and Chapter 4 (*Up Against the System*) has been accepted for publication. The listing of authors for *Innovations in Research with Medically Fragile Populations* is based on the considerable support and critical appraisal I received from Dr. Susan Jack in the development of a case study research design, guidance for analysis, a review of early coding to ensure rigor, and iterative feedback on the development of an innovative data collection strategy. In Chapter 4, Dr. Harold Siden is listed as the second author in recognition of his role as medical director at Canuck Place Children's Hospice, contribution to accessing this population of young adults with pedLTC, and support for partnering with this research study. Throughout the study, all committee members were instrumental offering guidance on study design, recruitment, analysis, and writing of the manuscript.

My contributions to the research include:

- Interest in the population of young adults with pedLTC transitioning from pediatric to adult services based on my experiences as a Clinical Nurse Specialist at Canuck Place Children's Hospice

- Application of qualitative methods acquired through previous experience, current courses, mentorship, and the mentorship of Drs. Susan Jack and Gina Browne.
- Design and implementation of the study:
 - Recruited study participants with assistance from Susan Poitras, Canuck Place Children’s Hospice counsellor and lead of the transition committee, who identified young adults fitting the study’s inclusion criteria.
 - Conducted all of the face-to-face interviews with transcription services from a professional transcriber.
 - Identified iTracks™ Bulletin Board Focus Group software, developed the Bulletin Board Discussion Group (with consultation from iTracks™ and an online bulletin board expert), tested the discussion board (with consultation from a young adult with pedLTC; my committee members; and transition, condition specific, and youth sexuality experts).
 - Conducted and moderated two – five day Bulletin Board Focus Groups. A professional transcriptionist transcribed video responses on the Bulletin Board Focus Groups. The Bulletin Board Focus Group discussion board was imported directly into NVivo9.
 - Coded all the face-to-face and Bulletin Board Focus Groups using NVivo 9™. I received two expert consultation sessions on the

management of data using NVivo™ from Dr. Guenther Krueger (NVivo™ consultant) and attended NVivo™ workshops.

- Prepared the original research protocol for ethics approval from two different ethics review boards: McMaster Health Sciences and the University of British Columbia. The Canuck Place Children's Hospice research committee also granted approval.

Finally, I wrote all of the manuscripts for this thesis. All manuscripts were reviewed by my committee members – Drs. Gina Browne, Susan Jack, Lehana Thabane, and Hal Siden; – who offered comments to a series of drafts that culminated in this final submitted version.

CHAPTER 1

INTRODUCTION

THE EXPERIENCES OF YOUNG ADULTS WITH PEDIATRIC LIFE
THREATENING CONDITIONS TRANSITIONING FROM PEDIATRIC TO
ADULT SERVICES:
A CASE STUDY

CHAPTER 1

THE EXPERIENCES OF YOUNG ADULTS WITH PEDIATRIC LIFE THREATENING CONDITIONS TRANSITIONING FROM PEDIATRIC TO ADULT SERVICES: A CASE STUDY

“Emily” is 20 years old and lives at home with her parents, sisters and two dogs. She attends university on a part time basis, makes cards to raise money for a charity, and likes music and online games. Sounds typical, but Emily’s life is anything but normal. She has a rare neuromuscular-type condition, still undiagnosed. She has never walked and was in her first powered wheel chair when she was 3 years old. After recurring chest infections, when she was 10 years old, she received a tracheostomy to provide a clear airway and mechanical support for her breathing. She is unable to swallow and is fed through a feeding tube. At age 20, Emily’s independent function is reliant on movement of her forehead and shoulders to activate button-like monitors that operate her power wheel chair and communication device. She “points” the button on her forehead at words, phrases and letters that appear as typed words on a computer screen attached to her chair. Her speech has regressed to vocalizations that her mom is adept at interpreting. She operates her power wheel chair via shoulder movements.

Physical limitations, the debilitating effects of her condition, and the equipment that surround Emily create a shield-like barrier that others find intimidating. Not surprisingly, Emily feels isolated and alone, detached from her peers and social networks. Yet, Emily is also resilient; she is hopeful, and she offers herself as a change agent to a world of people who cannot “see” her. This is her poem:

To Fit In

*I don't feel like I fit into the world
People don't see me because I am in a wheelchair
Only the people who know me see me as a person
I am the same as everyone else my age*

*Stop and say hi, I will smile and say hi back
But you won't understand my speech, so give me your email address and we can
chat
If you give me a chance, I can be a friend
I love to talk, but you have to patient as I type out my message on my lap top
Don't be afraid to talk to me*

*Little kids aren't afraid to talk to me
They ask 101 questions
Their parents worry about them asking the wrong questions, but I don't mind*

*I love a group where I can be myself
A group where I can be understood
Now that I have good friends, now I am happy.*

(Used with permission)

The following pages describe a case study that explores the experiences of young adults, like Emily, with pediatric life threatening conditions as they transition from pediatric to adult care services. Despite debilitating conditions imposed by their conditions, daily medical and exercise regimens, declining health conditions, and a limited horizon of opportunity, they persistently dream and aspire to

achieve academic goals, contribute to meaningful work, have intimate relationships and live independently. This study chronicles the health, education and social system barriers and facilitators they encounter in their young adult years, and presents examples of their resiliency, despite these barriers, to achieve their goals. “Emerging adulthood” (Arnett, 2000) is also an apt description of this age group and inclusive of their developmental goals, but focuses on the 18 – 25 year olds; whereas this study focuses on their experiences up to the third decade of life.

The Problem

Health and Technology Innovations and Advances

Advances in pediatric medicine and technology have created an unanticipated problem in adult services by transforming the outcomes of pediatric chronic conditions. Youth who previously would have not survived through adolescence are transitioning into an adult healthcare system that is poorly prepared to manage their complex pediatric conditions, and does not provide an equivalent level of services to support them into their adult years (Brown, Katherine, Allen, Quach, Chiu & Bialystok, 2010; Hamdani, Jetha & Norman, 2011; Stewart, Stavness, King, Antle & Law, 2006). It is estimated that 90% of children with special health care needs live beyond 20 years of age (Hagood, Lenker, & Thrasher, 2005) and that 70% of these children have four or more chronic illnesses (HSC Foundation, 2007). Embedded within this group of children with special needs, who may live for decades into their adult years, are

those with pediatric life limiting conditions (pedLTC) - such as Duchenne's Muscular Dystrophy (DMD) - who are living beyond childhood, but are not likely to live beyond early adulthood. For the purpose of this study, young adults with pedLTC are defined as those with progressive life limiting pediatric conditions, who have lived beyond their childhood years, but who are not expected to live beyond their first decade of adult life. The terminology used to describe young adults with pediatric life threatening conditions (pedLTC) varies. "Pediatric life limiting conditions" (pedLLC) and "pediatric life threatening diseases" (pedLTD) are also sometimes used. I chose to use pedLTC to distinguish this population from 1) pedLLC, which has been used to describe youth with conditions that may be life limiting, but with expectations to live well into their 50's and 60's, and from 2) pedLTD, which does not reflect that many of these young adults do not have a specific disease, but rather complex conditions with multi-system effect.

While there has been increasing interest in the transition from pediatric to adult health services over the past decade, there is little evidence of widespread implementation of health transition supports to achieve a basic standard of care (American Academy of Pediatrics, 2011; McDonagh, 2006). The continued development of professional and policy statements illustrates that recommended strategies have not been implemented (Berg Kelly, 2011). Young adults with pedLTC can be overwhelmed by the sudden decrease in services, from the plethora of integrated pediatric programs encompassing health, education and social services, to scant adult health services (Young, Barden, Mills, Burke, Law

& Boyell, 2009); and there is no evidence in the literature of transition services that will address their palliative care needs (Doug, Williams, Paul, Kelly, Petchey & Carter, 2011). Most young adults with pedLTC are not able to access developmentally appropriate palliative care services (iPANEL, 2011).

Transition: The Abyss

Health care transition is a complex phenomenon that is influenced by multiple and diverse factors (Wang, McGrath & Watts, 2010; Hamdani et al., 2011) such as providing skills and knowledge to youth and their families (Kennedy, Sloman, Douglass & Sawyer, 2007); training and coordination of health care providers in the pediatric and adult sectors (Lotstein, Ghandour, Cash, McGuire, Strickland & Newacheck, 2009); and family support (Baumbusch, Lynam, Sather & Wolverton, 2009; Stewart, Freeman, Law, Healy, Burke-Gaffney & Forhan, 2009). To date, the best examples of health care transition are confined to transfers between pediatric and adult subspecialty condition-specific clinics such as diabetes, cystic fibrosis and congenital heart defects and not complex chronic and progressively worsening conditions (Doug et al., 2011; Kennedy & Sawyer, 2008). Transition research has focused on providing specific transitional skills and medical knowledge to the youth and their family, as well as their health providers (Hamdani et al.). However, transitions from pediatric to adult care are more complex than condition-specific services; transitions are embedded within the larger health care system and interconnected to the education and social service

systems (de Camargo, 2011; Hamdani et al.). In 2009, the Deputy Chair of the Senate Committee on Population Health stated in the parliament of Canada that

Even if well integrated, healthcare services alone do not make a community healthy. Governments at all levels should support an overall, community-based approach to health and human development and assist with the integration of health and social services, wherever possible, and where it makes sense for the users (Nasmith et al., 2010, p. 14).

Further, health-promoting interventions are most effective if health services recognize and improve services for specific populations over their life course (Brown et al, 2010). The most effective strategies join health and social sectors together around a common set of goals that include the health, well-being and social context of children as they grow (Brown et al., 2010). Therefore, to achieve optimal care, an “interagency, trans-disciplinary approach” is required (de Camargo, 2011, p. 793).

In the United Kingdom, The National Health Service is integrating these systems into a “whole systems” approach (de Camargo, 2011). In British Columbia, (B.C.) Canada, some young adults with pedLTC have experienced a pediatric palliative care program in which the medical management of their condition, and their developmental, social, and educational needs have been integrated. Palliative care is an interdisciplinary approach to the management of incurable or life threatening diseases (Wein, Pery & Zer, 2010). When youth with pedLTC reach 18 years of age, appropriate adult health care services are difficult to access, and social and educational supports disappear just as they are experiencing profound life changes (Pinzon, 2009).

Research Study Background

I have worked with individuals and families living with chronic conditions and facing life-threatening conditions for most of my professional nursing career. In addition to working in this field as an advanced practice nurse, I have a designation in family counselling. This research study was borne out of a recent experience working in pediatric palliative care at Canuck Place Children's Hospice (CPCH). CPCH is a freestanding pediatric hospice, servicing the needs of children aged 0 to 19 years with progressive life-limiting conditions within the province of B.C.

Exemplary Model of Comprehensive and Integrated Services

The Canuck Place program offers comprehensive health, education and social services that include province-wide consultation, respite and family support, pain and symptom management, end of life care, psychosocial and spiritual support, consultative services, and bereavement care. This exemplary pediatric model illustrates the benefits of an integrated service program. CPCH creates an environment that supports a naturalistic way of life for children and youth through an onsite schoolroom, and enhancing quality of life through music, art, recreation, and cultural events.

Weekly multidisciplinary rounds are held at CPCH to discuss the health, social and educational resources required by the youth currently staying in the hospice. Starting around 15 years of age, or when developmentally appropriate, members of the Canuck Place team and transition committee provide individual

and group support to prepare the youth for transition to adult services, encouraging them to begin to take responsibility for their medical care and to investigate the services that they will need to attend post secondary institutions, create vocational opportunities, and live independently. Within a safe and mutually supportive environment, facilitated by a counsellor or social worker, youth explore physical development and losses related to their illnesses, emotions, independence, sexuality and social intimacy, mentorship, family relationships, survival and courage, and social support. “Teen camp” provides further opportunities for these youth to meet with their peers and young adults who have graduated from Canuck Place, to learn from their experiences and successful strategies to achieve their goals, and to discuss significant life and death decisions facing them as their condition progresses.

Close to their 18th birthday, and on their final day at Canuck Place, the graduates request their favourite items for the lunch menu. Small speeches and presentations are made, photos taken, and their friends and Canuck Place professionals and staff fete the graduate. Watching them on their final day with an appreciation for the community of support they are leaving, and the abyss of services lying ahead of them, is heart wrenching. There is no safe passage from Canuck Place to a similar supportive, holistic and comprehensive adult program. To an observer, it appears that these youth are wheeling into a void and uncertain future. While much has been written about the transition experience of youth with chronic health conditions, mostly from the perspective of health care providers

and parents, I was curious about the experiences of this vulnerable and medically fragile group of young adults from their perspective. Seeing so much to be accomplished in young adulthood, and an abbreviated time frame in which to accomplish those goals, I aspired to understand more about the opportunities and barriers they faced living their lives after comprehensive pediatric services, and in pursuing their passions and dreams.

Transition Outcomes for Young Adults after Pediatric Palliative Care

Young adults with pedLTC, who “graduate” from Canuck Place, do not have a safe passage to similar programs in adult services that might support them to meet their health, social, education and developmental needs. Instead, these young adults often become socially isolated due to their chronic health conditions. Increasingly dependent for some aspects of their daily living, they need to make psychological adjustments to their worsening condition and declining function (Fryer, 2004). Three of the most significant transition deficits for young adults with pedLTC are described here.

First, providing support for young adults with pedLTC is complex and requires a patchwork of services from health, human, social, spiritual and economic service providers (Nuffield Trust, 2010). In pediatric care, there is a growing body of clinical practice guidelines being developed for health care transitions in general, and for specific disease groups (Amara et al., 2011). However, these guidelines do not exist for the palliative population. Further, adult sector support services are underfunded compared to the pediatric population, and

coordinated services to provide seamless transition from pediatric to adult services do not exist (Amaria et al.).

Second, despite the advances made in pediatric palliative care in recent years, palliative care for young adults with pedLTC lags far behind the services provided to children. In B.C., three-quarters of adults with non-malignant diseases do not receive specialized palliative care services (iPANEL, 2011). Palliative and hospice services in adult health care are limited to the final few weeks or months of life (Birnkranz & Noritz, 2008), and consequently, are not available to young adults with pedLTC. In addition, these young adults are contending with the enormous psychological issues embedded in life limiting conditions and the impact of their condition on themselves and their family (Young et al., 2009), while striving toward “normal” developmental milestones within a compressed time frame (Meleis et al., 2000) without the support of expert palliative services. Therefore, the process of transferring youth with pedLTC from pediatric to adult services without palliative care is fraught with complications (Doug et al., 2011; Young et al.).

Third, the enormous amounts of time, money and resources provided to support and extend the lives of children with pedLTC abruptly end without consideration for how the intent to improve their quality of life will be extended into their adult years (Hamdani et al., 2011). Barriers to continuing coordinated services into adulthood increase the costs to the health care system, due to increased morbidity rates, resulting in more emergency room and hospital care

(Kennedy et al., 2007; Prestige, Romann, Djurdjev & Matsuda-Abedini, 2012; Rosen, Blum, Britto, Sawyer & Siegel, 2003). Integration among the young adults, their family, school and health and social service providers is missing in the transition of care for young adults with pedLTC.

Prevalence of Transition Concerns for Young adults with Pediatric Life Threatening Conditions

In B.C., nearly 4,000 children aged 4 and under have special needs (StatsCan, 2006). Canuck Place Children’s Hospice provides care to a select group of special needs children in B.C. with pedLTC such as severe prematurity, or life limiting neurological, metabolic and genetic conditions. There are currently about 175 children with pedLTC in the Canuck Place program, and every year 10-12 adolescents “graduate” from the program to adult services. However, the prevalence of young adults with palliative care needs is much higher than the mortality rate (Evans, 2011) because there are many more young adults in B.C. who will die of other pedLTC such as cardiac, neuromuscular, and oncological diseases. For example, 52% of cancer deaths and two thirds of deaths due to digestive and circulatory diseases are those of persons in their young adult years (Evans). The patient population of young adults with pedLTC will grow as earlier diagnosis and improved medical management lead to higher rates of survival.

Purpose, Research Questions, and Expected Outcomes

Purpose

The purpose of this descriptive case study was to explore the experience of transitioning from pediatric to adult services for youth with pedLTC. The specific aspects of the transitional process that are the focus of this study were:

1. Challenges and opportunities experienced by young adults with pedLTC leaving pediatric services and engaging in adult services and;
2. Young adult perceptions of the factors (individual, family, social and system-level) that influence the transition from pediatric to adult services

Research Questions

The research questions guiding this study were:

1. What are the experiences (opportunities and challenges) of young adults with pedLTC who have transitioned from pediatric to adult services?
2. What are the most significant supports and factors that will contribute to achieving their developmental goals?
3. What are the advantages and disadvantages of an online focus group designed to engage medically fragile participants with variable communication modalities and abilities, limited mobility, and who are geographically dispersed?

Expected Outcomes

The expected outcomes of this study were to:

1. Provide a comprehensive description of the challenges and opportunities experienced by young adults with pedLTC who have transitioned from pediatric to adult services.

2. Inform service providers and policy makers about health system and public policy innovations needed to support young adults with pedLTC transitioning from pediatric to adult care.
3. Contribute to foundational research in the emerging field of transitional care for youth and young adults with pedLTC.
4. Develop an innovative online focus group, and evaluate the merits of this data collection strategy with medically fragile participants who have variable communication modalities and abilities, limited mobility, and who are geographically dispersed.

Literature Search and Strategy

Strategy

A comprehensive literature search was conducted in 2010 and updated until May 2013. A search of Ovid, Medline, CINAHL and cited references (see Appendix A for key terms and results) was conducted to analyze journal articles related to the transition from pediatric to adult health services for the most prevalent causes of death from pedLTC in young adulthood (19-29 years) in B.C. While 27 articles were found, none of the articles addressed the transitioning from pediatric palliative care to adult palliative care, or for multiple complex chronic conditions such as degenerative neuromuscular diseases (Doug, Williams, Paul, Kelly, Petchey & Carter, 2011). A modest array of studies (five) explored transition from the perspectives of the young adults (non pedLTC) or their families or the opinions of health professionals. Of the few research studies

directed at health professionals, sample sizes were small and mostly referred to the opinions of pediatric, not adult health care professionals, who are key to transition success (Suris, Akre & Rutishauser, 2009).

Description of the Literature

The following review of the literature is complementary to the literature reviews provided in the upcoming manuscripts regarding an innovative method for accessing medically fragile populations, experiences of transition related to the health, education and social systems, and the resilient and transformative experiences of young adults with pedLTC. Specific research on youth and young adults with pedLTC is not available; therefore, other sources documenting transition experiences of youth with chronic health conditions were accessed.

The medical transition literature (of all types of illnesses and disabilities) is generally divided between studies that focus on specific conditions such as congenital heart defects, cystic fibrosis, rheumatoid arthritis, spina bifida, transplant recipients, and Type 1 diabetes, and studies that address broad transition issues applicable to most youth with chronic health conditions (Paone, 2006). The majority of these studies provide descriptions of transition experiences, expectations, needs and concerns, the transition process including program designs, and transition tools for readiness. However, these tools have not been validated, and there are few appropriate tools to assess the youth and family's readiness for transition (Sawicki, Lukens-Bull, Yin, Demars & Huang, 2011). While life-threatening conditions have some similarities to other chronic complex

conditions, the purpose of this review is not to review disease specific program design, transition tools, and programs aptly described elsewhere.

Further, there are very few outcome evaluations regarding transition programs that span both pediatric and adult services. An exception is a Canadian study in which it was found that even in a single disease specific program such as cardiology, where the complexity of transition variables are reduced because youth can be “transferred” from a pediatric cardiac specialist to an adult cardiac specialist, only 47% of patients transferred successfully to adult services, and 27% of young adults did not receive cardiac follow-up after leaving pediatric care (Reid et al., 2004). For young adults with pedLTC, the complexities of these problems burgeon where transfers from multiple pediatric subspecialists to adult specialists are required.

In addition to the literature describing disease-specific transitions, 18 references were found related to consensus and policy statements and general standards calling for organized, coordinated and efficient transitions for adolescents. Most of these models of care focus on the transition of adolescents with a specific condition, or the transition from a pediatric subspecialty to an adult subspecialty providers and focus exclusively on health but not developmental, educational and social needs. While there is agreement about the principles and models of effective transition, there is little outcome evidence to support these models (American Academy of Pediatrics, 2011; Lotstein et al., 2009; McDonagh, 2006). Overall, the literature is unanimous in its description of the transition

process as complex, multi-layered and multi-systemic involving the youth, his or her family, and primary and tertiary health and social services (Pinzon, 2009; Suris et al., 2009). However, the literature on the interplay of these multi-systemic barriers and facilitators for transition is limited (deCamargo, 2010; Hamdani et al., 2011).

The Role of Primary Care in Transition

Until recently, there has been little focus on primary care providers in the transition process (Betz, 2004). The role of a primary care medical home is now prominent in the clinical guidelines on transition from adolescence to adulthood as developed by The American Academy of Pediatrics (2011) and in a Canadian policy statement about child health systems caring for children with complex chronic health conditions (Brown et al., 2010). The philosophy of the medical home recognizes that transition and transfer are influenced by more than just medical care and include environmental, socioeconomic and other factors such as education, career choices and independent living. Interagency collaboration across services, interconnectedness between pediatric and adult services, and comprehensive transition practices and activities result in positive transitions (Berg Kelly, 2011; Brown et al.). A medical home establishes partnerships across programs and services, and with youth and their families, to provide explicitly and comprehensively planned care. Canuck Place Children's Hospice provides an "episodic" type of pediatric medical home where health, education and psychosocial services are provided "in house" and coordinated with community

agencies during the child's stay at the hospice. However, at the point of transfer, there is no equivalent service in the adult primary care or specialist sector to receive and continue to provide these services. Policies recommending a medical home are a substantive addition to the transition literature for youth with pedLTC, who are entering adult health care with complex lifelong histories, multiple medical interventions, and varying system navigation experiences (McAllister, 2011).

Criteria for Successful Transition

The American Academy of Pediatrics (2011), the Society for Adolescent Medicine (Rosen, Blum, Britto, Sawyer & Siegel, 2003) and the Canadian Pediatric Society (2007) have established principles for a successful transition to adult health care for adolescents and young adults with chronic health conditions. McDonagh (2006) provides a compilation of all of these principles that occur between 12-18 years of age and ideally transition:

- is an active, future-oriented process that is young person centered
- is inclusive of parents and caregivers
- includes age and developmentally appropriate education such as condition-specific information, life skills, self advocacy, peer and psychological support, educational and vocational planning, sexual health, drugs and alcohol information, and genetic counseling
- is multidisciplinary and interagency

- involves pediatric and adult services, in addition to primary care
- provides coordinated, uninterrupted health care that is age and developmentally appropriate, culturally appropriate, comprehensive, flexible, and responsive
- is holistic, including medical, psychosocial, and educational and vocational aspects (American Academy of Pediatrics, 2011; Canadian Pediatric Society, 2007; Green, Powers & Sells, 2003; Rosen et al., 2003; McDonagh, 2006; Sloman, 2005).

The Canadian Pediatric Society (2007) also stresses that health care goals for transition should include adolescent's involvement in the management of the condition(s), the adolescent and family's understanding of the condition, and the adolescent's realization of personal potential for activity, education, recreation and vocation, completion of adolescent developmental tasks and the attainment of self-esteem and confidence. Specifically in B.C., the provincial government has developed a Transition Planning Model for Youth with Special Needs, identifying best practices that might best support a person-centered approach to transition planning (Government of B.C., 2008). The ultimate goal of this model is to enable young adults with special needs to experience a self-directed life. The main principles of this model include a young adult-centered focus; young adult, family and community involvement; identification and use of a transition coordinator; and interagency collaboration.

Barriers to Successful Transition

Despite the abundance of guidelines, models, programs and consensus statements to improve the transition process from pediatric to adult services, many barriers remain. These include shifting from family-centered to patient-centered care, lack of resources, time frame for the provision of care, declining quality of life, and developmental factors. The current conditions that have created these barriers and the specific implications for youth with pedLTC will be described below.

Shifting from family-centered to patient-centered health care. The transfer from family-centered pediatric care to individual-focused adult care has been described by young people and their parents as “falling off a cliff” and “more stressful and difficult than anticipated” (Gorter, 2009; van Staa, Jedeloo, van Meeteren & Latour, 2011, p. 826). Pediatric care at its best is interdisciplinary, family-focused, developmentally appropriate, and considers the parents as team members in decision-making (Pinzon & Kaufman, 2007). Specialized multidisciplinary pediatric teams become a “medical home” for families, creating “accessible, family-centered, compassionate, continuous, comprehensive, coordinated and culturally effective” care (McAllister, Presler & Cooley, 2007, p. 724) that results in fewer medical errors and delays, and improved quality of life for the children and their families (Adams, Mahant & Cohen, 2009).

Rather than utilizing a team approach to care, adult practitioners focus on their specific specialties within a complex condition. Adult health care is patient-focused and investigational, requiring autonomous and independent decision-

making (Pinzon & Kaufman, 2007; Rosen, 1995). Without a team approach and a designated coordinator, communication among care providers becomes ineffective and increases stress for youth and their families (Adams et al., 2009; Young et al., 2009). Many youth with pedLTC are unable to be their own advocates and educators because of the complexity of their disease conditions, declining health and function, and dependence on medical technology. Ongoing support of family, peers, community members and pediatric service providers creates a safeguard for young adults in the transition process (Baumbush et al., 2009; Burke, Spoerri, Price, Cardosi & Flanagan, 2008; Lotstein, McPherson, Strickland & Newachek, 2005; Stewart et al., 2009). At the juncture when peer support is most crucial, youth with pedLTC become isolated when their pediatric hospice and high school communities end (S. Poitras, personal communication, June 13, 2013).

In addition to being under-resourced with experienced clinicians and multidisciplinary team coordination, many adult health care professionals and resources are available only in specialized tertiary care centers that may be very distant from the family's home (Burke et al., 2008; HSC Foundation, 2007; Wolfe, 2004). Further, communication and support between adult specialists and their primary care colleagues is compromised when there is no remuneration for time spent in telephone conferencing about specific patient needs. Therefore, limited interest in the transition of youth from pediatric palliative care to adult health care services, and inadequate services, capacity and knowledge about young adults

with pedLTC, heavily influence the experience of youth with pedLTC transitioning from pediatric to adult services (HSC Foundation, 2007).

Time frame of palliative services. The time frame allowed for hospice and palliative services in adult care is philosophically distinct from pediatric care. Both pediatric and adult palliative care services focus on relieving the physical, social, psychological and spiritual suffering experienced by the patient and family, and on quality of life. Pediatric hospice and palliative care applies these principles from the time of diagnosis through end of life, which may be years and years, whereas adult hospice and palliative care applies only to the last weeks and months of life (Fraser et al., 2012). Pediatric hospice provides both palliative and respite care over the child's lifetime, whereas adult palliative and respite care is only applicable within months of death.

This limited time frame for the provision of palliative and hospice services in the adult sector can prevent access to appropriate services for youth with pedLTC. The chronic trajectory of pedLTC often follows a series of declining plateaus in health status over a period of weeks to months to years, that are punctuated by periodic crises, and often a "sudden" death (Wolfe, 2004). The crisis events are complicated by their unpredictability, specifically regarding which one will be life ending. Therefore, the goals of care do not follow an "either/or" approach that focuses either on hope for extending life or on comfort. Rather, management of pedLTC requires treatment of the underlying condition

along with provisions for physical, emotional and spiritual needs simultaneously over unpredictable periods of time (Wolfe, 2004).

Impact of Pediatric Life Threatening Conditions on Quality of Life

Quality of life for young adults. Quality of life (QoL) studies have not been conducted on young adults with pedLTC or their parents and family members. Therefore, QoL studies for youth with complex chronic conditions and their families were reviewed to establish a baseline and to determine factors that may contribute to QoL for young adults. Almost all QoL studies in the pediatric literature focus on children with single chronic conditions (Arnaud et al., 2008; Klassen, Miller & Fine, 2004; Majnemar, Shevell, Rosenbaum, Law & Poulin, 2007; Varni, Limbers & Burwinkle, 2007), except for Thurston, Paul, Ye, Loney et al., (2010), who researched QoL outcomes for youth with complex chronic conditions. In this study, QoL scores for the youth decreased with advancing age, and QoL scores were negatively associated with the presence of depression and anxiety symptoms in the mother or primary caregiver. Further, despite ongoing physical and/or cognitive decline, youth with complex needs who scored higher on psychosocial well-being scales had lower pain levels, fewer behavioural difficulties, and lower parental distress (Thurston et al.). If extrapolated to young adults with pedLTC, QoL will decline with and be made worse by depression or anxiety in their caregivers. Also, QoL factors will become more significant as health and functionality decline during end of life trajectory. While QoL declines

for the young adults and their family, frequency and costs of health and social services will increase (Thurston, Paul, Ye et al.).

Quality of life for family. Family members' perspectives of the transition experience are not the focus of this research study, however, stresses experienced by the family affect the young adults and add a contextual richness. Parents are generally anxious about transfer of care and their diminishing role in health care decisions, and generally believe that adult practitioners are not knowledgeable about pediatric-onset conditions (Amaria et al., 2011; Reiss, Gordon & Walker, 2005). Family members of young adults with pedLTC may experience added stress and uncertainty, related to the increasingly complex medical, physical and psychosocial needs of their youth as they become young adults (Hatzmann, Maurice-Stam, Heymans, & Grootenhuis, 2009; Knafl, Knafl, Deatrck, Gallo, & Grey, 2006) while their system supports and resources significantly decline (Pinzon, 2009). Literature reporting on QoL for parents of youth with chronic illnesses will be used as a predictor of the escalating impact on families when supportive pediatric services end. Over 40% of Canadian parents of youth with multiple chronic and special needs conditions have symptoms of mild to severe depression (Thurston, Paul, Loney et al., 2010). In other studies, 29% of mothers are shown to have elevated symptoms of depression (Singer, 2006) and 12-15% have clinical depression (Bailey, Golden, Roberts & Ford, 2007). Both mild and severe depression are linked to detrimental individual, familial, and societal outcomes (Zimmer & Minkovitz, 2003), and even mild depression is related to

considerable personal limitations and distress in social, work-related, and physical functioning (Singer, 2006). Given the declining support in adult systems, and an increased burden of care as their young adult's health status declines, detrimental mental health outcomes can be expected for parents of young adults with pedLTC. Health and social service providers need to be cognizant of providing support for family members, to reduce, ameliorate and prevent escalation of these outcomes with attendant costs for services, as the youth's condition worsens (Singer, 2006).

In addition to the psychological burden of parenting a young adult with pedLTC, high-level skills are required to provide complex care, placing additional demands on family time and resources (Harrigan, Ratliffe, Patrinos & Tse, 2002; Rehm & Bradley, 2005). Many young adults require ongoing skilled and supportive care that includes assistance with all aspects of daily living (getting up, dressed, bathing, feeding), assistive technology, complicated dietary regimens, and physio and occupational therapy (Rehm & Bradley). Further, emotional and financial stresses accrue as end of life approaches (Harrigan et al.; Wang & Barnard, 2004). Effective intervention for these families must include QoL components that include education, emotional support, skilled supportive and respite care, and financial assistance (Harrigan et al.; Wang & Barnard).

Developmental Factors

For young adults with pediatric life threatening conditions.

Developmental issues that impact the success of a youth's transition to adult services and ability to overcome system barriers as described above, will be

summarized here. The transition to adult services is a tumultuous time of upheaval and change for young adults. Biological, social, developmental and cognitive changes are inherent in the young adult stage (Suris, Michaud & Vine, 2004). Developing the maturity to participate in adult services impacts the success of transition from pediatric to adult care (Lotstein, et al., 2005; Burke et al., 2008). In the pediatric system, care plans and strategies are built around a young patient's developmental stage, ensuring that the child receives the care he needs regardless of his chronological age. The adult health care system is based only on the expectation that an 18 year-old is an adult. However, the prognosis of a pedLTC, repeated and prolonged hospital stays, intrusive home care regimens, sporadic school attendance, and fewer socialization opportunities delay some youths' development of independence and autonomy, including the requisite skills to engage in the adult health care system (Fryer, 2004). The inclusiveness of family-centered care can result in youth always being seen by the health care team with parent present and precluding opportunities to develop self-management skills (Amaria et al., 2011). Further, even with normal developmental opportunities, full adult functioning brain capacity does not occur until the mid twenties (Steinberg, 2010). At 18 years of age when transition occurs, the brain does not have the capacity to engage in sophisticated planning and problem solving skills, both of which are requisite to successful adaption to adult services, even barring delayed development (Simpson, 2008; Steinberg, 2010). Finally, a study to determine the impact of pediatric chronic and life threatening conditions on life course and

transition into adulthood found that all these young adults achieved fewer milestones, or at an older age than their peers, and those with a pedLTC were affected the most (Stam, Hartman, Deurloo, Groothoff & Grootenhuis, 2006). Health and other service providers can encourage parents to foster social contacts and autonomy to mitigate the effects of living with complex conditions.

For their parents. After nearly two decades of managing the complexities of their child's health condition and systems providers, and being integral to decision making, relinquishing these responsibilities is challenging for parents (Rosen et al., 2003; van Staa et al., 2011). Striking a balance between relinquishing management and remaining supportive of their young adult in assuming some of these responsibilities is important. Current initiatives in clinical practice and research call for youth to become increasingly involved in decision-making, and yet parental involvement in transition increases successful outcomes (American Academy of Pediatrics, 2002; Betz, 2004; Government of B.C., 2008). Further, parents of young adults with pedLTC may anticipate an uncertain and brief time frame in their relinquished role prior to becoming responsible again when their young adult's health status declines.

Conceptual Foundation of Transition for Young Adults with Pediatric Life Threatening Conditions

As described above, little attention is focused on the transition experiences of young adults with pedLTC. In addition to the complexity of their condition, the fragility of their health status, and the enormous developmental changes they are

experiencing at the brink of adulthood, the superimposed losses associated with changing health, social and educational services and providers elevates the vulnerability of young adults with pedLTC during this transition (Fryer, 2004; Meleis et al., 2000). It is important to note that it is not necessarily the health condition that creates vulnerability, but the questionable availability of equivalent supportive services and resources (Suris et al., 2004; Ungar, Brown, Liebenberg, Cheung & Levine, 2008). Navigating through uncoordinated programs and negotiating for services, so that young adult goals might be achieved in an abbreviated time frame requires monumental effort and investment. From my pre study observations and reflections of this population, transition, growth and development, grief, and vulnerability were inclusive in their transition experience from pediatric to adult services and important to consider in the development of study questions, purpose and outcomes.

Development and Transition

During transition, youth with pedLTC simultaneously experience developmental and health, educational, and social system changes while situational and organizational supports also change (Meleis et al., 2000). The complexities of these interacting transitional components are further influenced by the following developmental factors: their awareness and willingness to engage in the transition process, their preparation, knowledge, beliefs and attitudes, and their maturational level (versus age) (Burke et al., 2008; Lotstein et al., 2009; Meleis et al.). Examples of engagement for youth with pedLTC include

seeking out information about suitable community resources for social support, becoming an active participant in their interactions with health care professionals (Reiss et al., 2005), and determining the accessibility and flexibility of post-secondary institutions. The degree to which youth with pedLTC will actively seek out developmental milestones such as intimacy, independence and the achievement of vocational or educational goals will be largely determined by their health status and the social, personal, environmental and educational supports available to them.

Development and Loss

Adolescence is a time of both enormous physical changes and the advent of growing independence, social skills, self-esteem and awareness. For young people with pedLTC, adolescence and early adulthood is “a paradox of emerging capabilities and diminishing possibilities” (Fryer, 2004, p. 381). Physical changes normally associated with puberty may be delayed and physical capabilities limited, due to weight loss, muscle weakness, and profound fatigue (Fryer). Further to these unwelcome physical changes, the impact of the pedLTC and its treatments may interfere with critical life experiences that are necessary to achieve the normal developmental goals of adolescence and young adulthood (Fryer). Developmental challenges for young adults with pedLTC emerging into adulthood include being less mature than their peers because of parental overprotection and learned passivity, feeling isolated from peer groups, and difficulty participating in social, athletic and intellectual activities (Fryer).

Independence and self confidence may be thwarted due to fewer opportunities to separate from parents, integrate into peer groups, and pursue post secondary-education and vocation (Fryer). Further, the joy of experiencing developmental milestones such as intimacy and independence may be concurrent with declining health and function. As Figure 1 demonstrates, for many types of pedLTC, the end of life trajectory is marked by a slow decline with periodic crises and “sudden” death (Wolfe, 2004); however, their desired goals and ambitions will be on an upward trajectory.

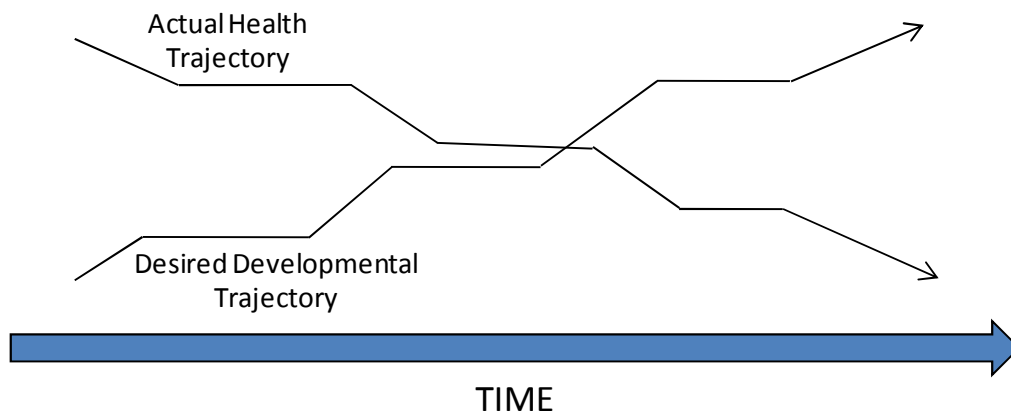


Figure 1. Health Trajectory (adapted from Wolfe, 2004) versus Desired Developmental Goal Trajectory

The growth and change process of young adulthood is described as being “alternatively exhilarating, disconcerting, satisfying, frustrating, and terrifying” (Simpson, 2008, p. 1). The vulnerabilities and the objective complexities of living with a pedLTC, along with declining physical function, increasing symptoms, periodic health crises and decreasing support from health, social and educational services, could also thwart some youth from realizing their “hoped for” goals.

However, personal beliefs and attitude, and the environmental supports that include family, friends, social and educational services, can have an enormous impact on a young adult's ability to reframe these potential obstacles. Clinical and anecdotal experiences demonstrate the capacity of these youth to maintain their vitality and zest for life and their perseverance in achieving their goals until the very end of their days.

Vulnerability

Rogers (1997) demonstrates that vulnerability is the result of the interaction between personal resources (cognitive, emotional, intellectual and behavioural) and environmental supports (social, material and cultural). In youth with pedLTC, vulnerability may be higher due to the long-term effects of their complex chronic illnesses and medications, and inadequate environmental supports from health, social and educational services. Conversely, growing up with a complex chronic condition requiring frequent contact with the health care system, missed school, and experiences that are widely divergent from other children their age may also develop resilience and strength (Calhoun & Tedeschi, 2006) that lowers their vulnerability.

Specific examples of personal factors that vary amongst young adults with pedLTC include their cognitive capacity, temperament, self direction, imagination, values and motives, and parental influences, such as their acceptance of the young adult's decision making, attitudes or preoccupation with their strengths or limits, and their view of the impact of the pedLTC on siblings and family. In addition to

the health, education and social systems, environmental supports include parents, siblings, extended family, teachers, friends, spiritual and cultural mentors, and volunteers.

Figure 2 demonstrates that the degree of vulnerability experienced is related to the quality of personal resources and the environmental supports available. The base of the triangle represents the vulnerability index (Rogers, 1997) when extrapolated to health and social services, it can be expected that increasing vulnerability can be associated with increased use and thus cost of these services (Markle-Reid, 2002).

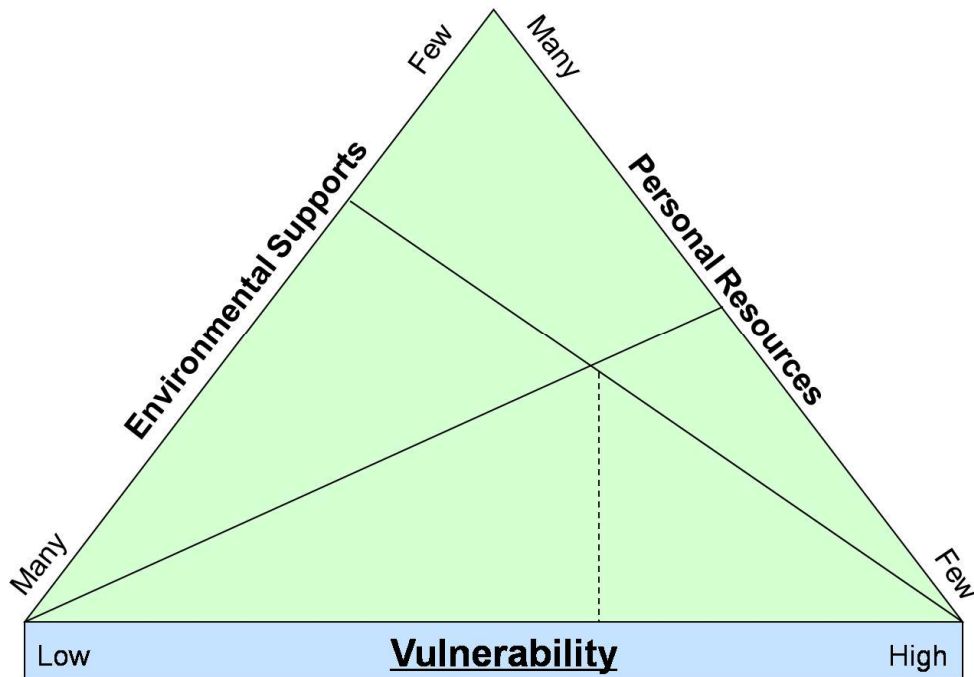


Figure 2. Degree of Vulnerability in Relation to Environmental Supports and Personal Resources (adapted from Rogers, 1997 in Markle-Reid, 2002).

To estimate an individual's degree of vulnerability, one would locate the person's level of environmental supports on one side of the triangle and

draw a straight line from this point to the opposite apex. The same would be done for the person's personal resources. At the point where the two lines intersect, a line would be dropped perpendicular to the base to intersect the vulnerability continuum. The point of intersection, the degree of vulnerability, therefore, represents an outcome of the interaction of personal resources and environmental supports (Rogers, 1997, p. 68).

As Figure 2 (Rogers, 1997) demonstrates, even if personal resources hold constant, changes in the individual's environmental supports can significantly alter vulnerability. For young adults with pedLTC, increasing vulnerability occurs after transition, when comprehensive pediatric resources cease, and equivalent resources in the adult system are unavailable or difficult to attain. Subsequent lack of engagement with the health care system threatens their adherence to treatment and regimens, resulting in increased morbidity and costs in acute care settings during crises (Kennedy & Sawyer, 2008). Inability to locate supportive resources in the education and social support systems will delay or preclude their opportunities to achieve post secondary and vocational goals.

Thesis Outline Overview

The remaining chapters of this dissertation are comprised as follows. Chapters 2 and 3 describe the application of case study methods used in this study. Chapters 4 and 5 present the results of the study, and Chapter 6 provides an overview of the research and a final discussion.

Chapter 2

Chapter 2 provides a comprehensive description of the case study that includes: type of case study and rationale, propositions that bind the case,

philosophical stance, sampling, recruitment and data collection, analysis, trustworthiness and ethical considerations.

Chapter 3

Chapter 3 addresses research question #3:

What are the advantages and disadvantages of an online focus group to engage medically fragile participants with variable communication modalities and abilities, limited mobility, and who are geographically dispersed?

This paper has been submitted for publication and follows the formatting style of the journal. “Innovations in Research with Medically Fragile Population’s” provides a detailed description of the development of an innovative online data collection strategy, the Bulletin Board Focus Group. An electronic link to view the discussion board set up for this study is provided. In addition to the description of the methods provided in this chapter, an abbreviated methods section, suitable for publication of manuscripts, is included in each of the articles in Chapters 4 and 5. Repetition of methodological, and elements of the study background are unavoidable.

Chapters 4 and 5

Chapters 4 and 5 respond to research questions # 1 and 2:

1. What are the experiences (opportunities and challenges) of young adults with pedLTC who have transitioned from pediatric to adult services?

2. What are the most significant supports and factors that will contribute to achieving their developmental goals?

Chapter 4, “Up Against the System”, describes barriers and facilitators in the health, education and social systems that the young adults with pedLTC experienced during their transition years. Despite the shortcomings of system and environmental supports, and the burden and impact of their conditions, these young adults demonstrated uncharacteristic resilience and hopefulness.

Chapter 5, “Investing in the Future”, explores the concepts of resiliency, vulnerability, and future thinking; it describes how young adults with pedLTC invest in their short future, and it provides examples of environmental supports and barriers in their quest to dream and achieve their goals.

Chapter 6

Chapter 6 summarizes key elements of Chapters 2, 3, 4 and 5, and provides a final discussion of the overall study purpose: To explore the experience of transitioning from pediatric to adult services for youth with pedLTC, with a specific focus on 1) the challenges and opportunities experienced by young adults with pedLTC leaving pediatric services and engaging in adult services, and 2) young adult perceptions of the factors (individual, family, social and system-level) that influence the transition from pediatric to adult services. Final analysis of the use of an innovative online strategy to collect data will also be presented. Findings from this study have expanded the theoretical underpinnings of the study offered in the conceptual

framework described in Chapter 1, and an adaption of this initial framework is provided. A summary of the strengths and limitations of this study, a reflection concerning future research, and implications for practice completes this dissertation.

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CHAPTER 2

METHODS

OVERVIEW OF THE CASE STUDY

METHODS: OVERVIEW OF THE CASE STUDY

Rationale for Case Study Design

A recent Delphi survey of Canadian clinicians and academics determined that not enough is known from the perspective of young adults about what they believe they need to make a successful transition from pediatric to adult care (Fletcher-Johnson, Marshall & Straatman, 2011). Round table sessions and policy have developed the best practices for transition among health care providers, but very little research specifically examines the perspective of the young adult who has experienced the transition process (Fletcher-Johnston et al.), and none examines the perspective of young adults with pediatric life threatening conditions (pedLTC) who are transitioning from pediatric palliative care services. A qualitative case study was chosen to facilitate an in depth exploration of the transition experiences for young adults with pediatric life limiting conditions (Yin, 2009).

Type of Case Study

I chose Yin's (2009) criteria for building a case study to explore the experiences of young adults with pedLTC transitioning from pediatric to adult services. Specifically, a descriptive single case study design with embedded units was selected to examine the complexity of the transition process for young adults with pedLTC. Yin describes the revelatory nature of the case as one of several rationales for conducting a single case study. This case is revelatory because these young adults represent a new and unknown cohort. Until recently, most would

have died before reaching adulthood. Case study supports the exploration of their pioneering efforts into health, education and social systems that are ill equipped to meet their needs.

This single case study investigated one issue (the transition experience from pediatric palliative care to adult care) embedded within the perspectives of a varied group of young adults with pedLTC (Baxter & Jack, 2008). Embedded units within the larger case study created the opportunity to analyze the data within each of the young adults (subunits), between young adults, and across all young adults (cross-case analysis) (Baxter & Jack). Among the many different types of case studies, a descriptive case study was chosen because this study describes the complex factors experienced by young adults inherent in their transition to adult services and incorporates a variety of data sources and collection methods (Baxter & Jack; Yin, 2009). Further, case study facilitates an investigation of an experience that lacks discrete boundaries around the experiences, uses multiple data sources, incorporates several theoretical perspectives, and recommends change-focused improvements (Yin). These factors are described below.

Complexity. In this case, the complex phenomena and their context included young adults with pedLTC:

- who have had a progressive pediatric life threatening illness most of their lives, and though becoming adults in age, may not necessarily

have the developmental ability or experience to participate in adult-oriented spheres of self-care;

- who are leaving long term and trusting relationships with pediatric health, education and social services workers who (at least in part) have understood the complexity and implications of their health condition on their development;
- with complex health and social needs transitioning from the context of comprehensive pediatric services, where health, social and educational needs are integrated, to adult systems of care where there is no integration among health, education and social service sectors; and,
- simultaneously experiencing two opposing forces: desiring to achieve developmental milestones (such as educational and vocational goals, and independence) while experiencing or anticipating declining health and function.

Multiple data sources. In addition to the capacity of case study to encompass the complexity of the topic, it also facilitates investigating the phenomenon from the perspectives of multiple data sources. For this study, multiple data sources (Yin, 2009) included interviews with young adults and expert consultants, such as health and social service providers; a review and analysis of documents, such as practice reviews; recommendations and policies; and my own clinical observations. Combining multiple data sources with theories of transition, vulnerability, growth and development, and loss and grief, ensures

that the phenomenon is explored through a variety of lenses, enhancing the trustworthiness of the data (Baxter & Jack, 2008; Eisenhardt, 2002; Yin).

Blurred boundaries. Case study methods are also well suited to situations in which boundaries between phenomena and context can become blurred. For example, in this case study, the phenomena of the young adults' transition experience from pediatric to adult services cannot be separated from their context, which includes the impact of their health condition; personal factors; and environmental supports such as their community and the health, education and social systems.

Transformative and change focused. Finally, the findings of a case study can be effective to change policy and practice, and promote more effective supports and services for young adults with pedLTC (Institute for Health Care Improvement, 2009).

Philosophical Stance

Philosophically, case study is categorized within the constructivist paradigm, recognizing the importance of both subjective and objective perspectives to create meaning (Baxter & Jack, 2008; Creswell & Plano, 2007;). Both social constructs of meaning created from the data, and the "*a priori*" theories and conceptual frameworks described in Chapter 1, can be encompassed within this paradigm. Further, theoretical propositions shift the philosophical stance towards the critical paradigm that straddles constructivist and post-positivist philosophies (Doyle, Brady & Byrne, 2009). This dual view allowed me

to be responsive to new ways of thinking about issues that arose during the research process, and to collect and integrate data from a variety of sources (Baxter & Jack; Eishenhardt, 2002; Yin, 2009).

Study Questions, Expected Outcomes, and Propositions

Study Questions

The questions guiding this case study were:

1. What are the experiences (opportunities and challenges) of young adults with pedLTC who have transitioned from pediatric to adult services?
2. What are the most significant supports and factors that will contribute to their achieving their developmental goals?
3. What are the advantages and disadvantages of an online focus group designed to engage medically fragile participants with variable communication modalities and abilities, limited mobility, and who are geographically dispersed?

Expected Outcomes

The expected outcomes of this case study were to:

1. Provide a comprehensive description of the challenges and opportunities experienced by young adults with pedLTC who have transitioned from pediatric to adult services.
2. Inform service providers and policy makers about health system and public policy innovations needed to support young adults with pedLTC transitioning from pediatric to adult care.

3. Contribute to foundational research in the emerging field of transitional care for young adults with pedLTC.
4. Develop an innovative online focus group, and evaluate the merits of this data collection strategy with medically fragile participants who have variable communication modalities and abilities, limited mobility, and who are geographically dispersed.

Propositions

Propositions were established prior to data collection to focus and bind the scope of the research questions, and to suggest possible links among the phenomena being studied (Yin, 2009). These propositions were derived from a review of the literature, my professional experience, and the opinions of experts in the field. These experts include adolescent, pediatric palliative care, neuromuscular and transition clinicians. The theoretical underpinnings (development and transition, development and loss, and vulnerability) of these propositions were described in Chapter 1. The propositions for this study included:

1. Development of comparable adult medical and support services did not follow interventions to increase life expectancy (Brown, Katherine, Allen, Quach, Chiu & Bialystok, 2010; Hamdani, Jetha & Norman, 2011).
2. Health care transitions are embedded within the broader health care system and connected to the family, social, educational, cultural, religious and many other systems (Brown et al.; Hamdani et al.).

3. Young adults with pedLTC are potentially juxtaposed between the hope of achieving new young adult aspirations and their own end of life trajectory (Doug, Williams, Paul, Kelly, Petchey & Carter, 2011; Freyer, 2004; Melies, Sawyer, Im, Hilfinger & Schumacher, 2000).
4. A continuation of the integrated palliative care services (including medical, psychosocial, educational, spiritual and social needs) received from 0-18 years is not available to young adults with pedLTC in British Columbia, and this unavailability could result in increased morbidity, isolation, and inadequate support for fulfilling their young adult aspirations for intimacy, independence, education, work and interpersonal relationships (Meleis et al.; Prestige, Romann, Djurdjev & Matsuda-Abedini, 2012).
5. Health and social service providers in the adult care sector could have limited awareness of the complexity of the issues for young adults with pedLTC, or do not have a mandate, or the resources to provide integrated services to young adults with pedLTC in B.C.
6. Current transitional services from integrated pediatric services to the adult health and social service sectors are limited in their breadth, scope and effectiveness to meet the needs of young adults with pedLTC (American Academy of Pediatrics, 2011; McDonagh, 2006).
7. Vulnerability; personal factors such as cognitive capacity, temperament, self direction, imagination, values, and meaning of transitional phase;

parental influences such as availability, history of acceptance of young adults' decision making, attitudes or preoccupation with young adults' strengths or limits, view of impact of young adults' pedLTC on siblings and family; and external supports such as extended family, teachers, community, religious, and cultural mentors can all influence the young adults' experience of the transition process (Suris, Michaud & Viner, 2004; Ungar, Brown, Liebenberg, Cheung & Levine, 2008).

Data Sources and Sampling

Data Sources

Following the best principles of case study research, triangulation of data sources was paramount to ensure completeness, enhance trustworthiness and confirm data findings (Creswell, 2007; Yin, 2009). A variety of data sources, including young adults with pedLTC, relevant clinical experts and service providers, literature on program development for young adults with pedLTC in the United Kingdom, and relevant policies and recommendations were accessed for this study (Eisenhardt, 2002; Yin). Further, two different types of interview methods, face-to-face interviews and an asynchronous online focus group were employed with the young adults to enhance the quality of the data and facilitate a deeper and richer understanding of their experiences over time (Yin). All other interviews with clinical experts and service providers were face-to-face or via telephone.

The quality and depth of the data I was able to glean from the research

interviews was enhanced through my work as a project manager for a Youth Transitions project, funded by the British Columbia Medical Association and affiliated with British Columbia's Children's Hospital. In this role, I was actively interviewing and collaborating with medical and allied health clinicians in pediatric subspecialties, as well as adult specialists, general practitioners and social service providers, about their perceptions of the transitional barriers and solutions to improve the transition process for youth with chronic health conditions. In addition, I facilitated three advisory meetings that convened transition experts from medical, allied health, and social sectors from across British Columbia, and coordinated a workshop on youth transitions for the British Columbia Medical Association's Family Practice annual meetings. My experience and immersion in these work opportunities informed the data collected through this study.

Sample: Young Adults

Purposeful sampling was chosen to demonstrate differing perspectives of the accessible populations of young adults with pedLTC (Creswell, 2007; Miles & Huberman, 1994). In this study, a purposeful sample of young adults with pedLTC, between 19 and 29 years, with limited expectations to live beyond their first decade of adult life (29 years), were selected from young adults who graduated from Canuck Place Children's Hospice (CPCH) with at least three visits in their teen years, and at least one of the following conditions:

- Circulatory system (congenital heart defects)

- Neuromuscular (Duchennes Muscular Dystrophy, Spinal Muscular Atrophy)
- Metabolic conditions (excluding PKU)
- Neurological
- Undiagnosed

Table 1 describes the prevalence and rate of death for these and other disease groups in young adults in B.C.

Table 1

Prevalence of Death for Young Adults (20 – 29 years) with Pediatric Life Threatening Conditions in British Columbia, (adapted from Evans, 2011, p. 101)

Condition	# of deaths	Total # of cases	% of deaths in young adulthood years
Neoplasms	128	246	52
Endocrine, nutritional and metabolic diseases	28	75	38
Diseases of the nervous system	45	104	43
Diseases of the circulatory system	69	100	69
Diseases of the respiratory system	26	61	43
Diseases of the digestive system	28	45	62
Congenital malformations, deformations and chromosomal abnormalities	31	207	15

Within the hospice, the numbers of young adults with pedLTC transitioning from pediatric hospice are very small (8 -10 per year) and only some of these graduates met the following selection criteria: English speaking, cognitively able to participate in their care and decision-making, owning a computer, and having

either verbal or typing ability sufficient to participate in face-to-face interviews or an online focus group developed by the researcher.

CPOCH has transitioned approximately 97 young adults from its program in the past 15 years. Of these, approximately half (45) were eligible for this study (N. Chavosi, personal communication, June 10, 2013). Of these participants, the number of young adults with pedLTC was limited to those that the CPOCH transition coordinator assessed would be suitable to participate in interviews, and for whom current contact information was available. Further, sample size was limited to the capacity of two BBFGS (12 participants), plus the face-to-face participants. Most of these young adults require 24-hour attendants for personal, respiratory, and feeding support. Their independent function may be limited to minimal movement of their index finger or a dot on their forehead to control their power wheel chairs, computers and phones. All the participants have lived with their conditions for more than 10 years, and most were diagnosed when they were less than 5 years old.

Sample: Specialist Informants

While the purpose of this study was to understand the transition experience from the perspective of the young adults, other informants were selected to participate in an interview when information gaps arose that the face-to-face participants were unable to explain. The following informants were selected: a neuromuscular nurse clinician, a post doctoral fellow specializing in the sexuality of young adults with disabilities, a family practitioner managing the

care of a young adult with a pedLTC, and a social housing resource allocation provider (who did not respond to requests for an interview).

Sample: Documents

With increasing awareness of the difficulties experienced by youth with chronic and complex health conditions transitioning from pediatric to adult services, many professional and health organizations, advocacy groups, and governmental and non governmental organizations have prepared documents outlining the current problems with transition within their jurisdictions and recommendations for change. In the United Kingdom, some pediatric palliative care advocacy groups have started to lobby governmental representatives to ensure that care provisions provided in pediatric palliative care are carried through to adult services. I sampled and reviewed documents that were written to describe: 1) the young adults' experiences leaving pediatric palliative care programs in Britain, 2) the development of young adult hospice programs in Britain, 3) innovative strategies to create integrated and accountable programs of care, and 4) newsletters and email circulars among clinicians and organizations working to bridge the gap in services for young adults with pedLTC. Documents were chosen for their relevance to young adults developmental issues, and their experiences of the barriers and facilitators to health, education and social services systems complicated by disabilities or complex or palliative conditions.

Recruitment

CPCH requests all families to consent to receive information about applicable research opportunities and maintains records of those who have transitioned from the program. The teen transition coordinator selected participants who were able to articulate their experiences in either a face-to-face interview or online focus group format. A purposeful sample of individuals who met the inclusion criteria stated above received a letter from the Medical Director of CPCH, informing them of an opportunity to participate in a research project about their experiences transitioning to adult services. The recruitment and consent letters (Appendices B and C) were sent out in two phases: first for the face-to-face interviews, and second for the Bulletin Board Focus Groups (BBFG), an asynchronous online focus group. The letters included details about the purpose, risks and benefits and confidentiality of the study. If interested, the young adults were asked to contact the researcher via email or telephone. The researcher emailed or phoned the interested young adults to provide further details of the study, and assess their eligibility to participate.

In the first phase, the first two CPCH graduates contacted agreed to participate in the face-to-face interviews. Consent forms were completed at the first interview. In the second phase, of the 12 young adults meeting the eligibility criteria described above, eight agreed to participate. Electronic copies of the consent form were sent to each participant's email address. After reviewing the consent, the participant typed or signed his or her name in the signature box, and returned the consent to the researcher via email or fax, prior to the commencement of the

BBFG. After three reminders to return the completed consent forms via email, fax or mail, if the participant joined the BBFG discussion, their participation was taken as consent.

Other specialist informants, such as a nurse clinician expert in neuromuscular diseases, a social service housing expert, and a post-doctoral fellow studying the sexuality of youth and young adults with disabilities, were recruited to fill in knowledge gaps arising from the participant interviews. These participants were recruited through email (Appendix D).

Data Collection

Development of Data Collection Tools

Face-to-face interview guides. The face-to-face interview guide (Appendix E) for young adult participants was developed from a review of: 1) the literature about transition experiences of youth with complex health conditions, 2) a video “All About Change,” (Merwitzer, 2010), made by a group of British young adults with life threatening conditions who have transitioned from pediatric to adults services, 3) input from pediatric palliative care transition team members, 4) a background interview with a young adult living with a pedLTC who exceeds the age limit for this study, and 5) recommendations from pediatric palliative care and young adult hospice advocates in the United Kingdom.

Bulletin board focus groups. The development of the BBFG is fully described in the next paper, “Innovations in Research with Medically Fragile Populations”. Briefly, an analysis of the face-to-face interviews, in addition to all

of the resources listed above in the development of the face-to-face interviews, supported content development of the BBFG.

Face-to-Face Interviews

In the first stage of this case study, two young adults with diverse and unique experiences that differed regarding characteristics such as diagnosis, age, gender, family support, opportunities for post secondary education and vocation, and living arrangements, were invited to participate in three face-to-face interviews, each lasting 30-60 minutes. These participants were similar in their physical limitations, requiring 24-hour care and possessing minimal movement adequate to control their power wheel chairs, computers and phones, but varied widely in terms of significant variables from the literature related to successful transition outcomes, such as satisfaction with present condition; connectedness and support from family; and social, health, and community networks. One participant was able to speak directly with me, and the other used a computer-assisted device to type out answers.

I invited the two young adults to meet with me at a convenient, private and comfortable place to conduct their individual interviews. Each was individually interviewed three times to ensure a thorough investigation and understanding of his or her transition experiences, build on the proposed theories and propositions, discover new emerging theories, and clarify and contrast with what is known in the field (Yin, 2009). Each interview was limited to less than an hour to ensure that the experience was not unduly tiring. While no significant unwanted effects

were expected, they were provided with the contact information for a teen counselor known to them.

A semi-structured interview format was followed based on the review of the literature and propositions, input from pediatric palliative care specialist clinicians who have facilitated teen and transition support groups, and the professional experience of the investigator. Questions used to guide the conversation in an unbiased and conversational manner are included in Appendix E.

With participant consent, interviews were recorded with a digital recorder and transcribed verbatim (Creswell, 2007). Field notes and observations, and thoughts about emerging concepts were documented immediately after the interview (Jack, 2005). The following table details the face-to-face interview protocol.

Table 2

Face-to-Face Interview Protocol

Procedures	Specifics
<i>Pre Interviews</i>	
1. Identify 2 interviewees with children’s hospice transition coordinator	Purposeful sampling to achieve a range of characteristics that may influence transition: gender, age, diagnosis, family support, opportunities for post-secondary education and vocation, and living arrangements
2. Send out recruitment letters via email and request participants to respond to researcher via email to indicate interest (or not). Reminder follow-up by hospice transition coordinator	Provide introductory information about the study: background information about the study, length and number of interviews, request to transcribe the interviews, and consent form
3. Contact participants via email or phone (depending on preference)	One participant preferred email correspondence; the other

to set up first interview	participant's mother communicated via telephone
4. Prepare and test recording equipment	Set up separate recording files for each participant
5. Prepare labelled and numbered folders for each participant's study documentation	For confidentiality and study administration, create a unique ID code for each interview and corresponding documents and consent forms.
First Interview	
6. Introductions, discuss study and background information, and request permission to record interview. Sign consent forms: study participation, permission to record and transcribe interviews, and requests for further contact for clarification or additional questions (if they arise) after the study.	Interviews conducted in person with participants at a location of their choice (their homes). Review purpose of the study, what to expect, risks and benefits, contact information for counsellor, and confidentiality of the study. Provide copies of study background information and consent forms to participants: 1 copy for researcher and 1 copy for participants.
All Interviews	
7. Conduct the interviews	Conduct 3 interviews (each 30 – 60 minutes) with each participant, using the interview questions as a guide to direct questioning and ensure all planned content is covered. The pre-planned interview questions are just a guide. Researcher listened carefully to participants to follow their direction.
Post Interviews	
8. Management of interview content	Electronic interview recording: <ul style="list-style-type: none"> • Each interview electronically dated and identified by unique interviewee code. • Interviews downloaded from recorder to computer via electronic files. • Electronic files sent to professional transcriber and returned to researcher via password protected drop box file.

	<ul style="list-style-type: none">• Electronic recording files erased.• Ensure transcribed copies are clean, remove identifying information• Hard copies of transcripts stored in locked cupboard• Electronic copies stored on password protected computer• Transcripts transferred to NVivo 9™
9. Write up contextual memos and audit trail	Memos of each interview completed immediately after the interview. Memos consisted of: <ul style="list-style-type: none">• Memorable interview highlights and challenges• Personal reflections of the experience, interview successes and challenges• Questions and hunches for follow-up in the subsequent interviews with the interviewee and other participants• Comparing and contrasting experiences of both participants with each other and literature about transition
10. Letter of thank you and gift card of choice given to each participant	

The following field note entry reflects my impressions of the personal and contextual differences of my face-to-face participants.

Interviewee 1

I arrive to a low-slung 60's ish bungalow style institutional building. Paint, repairs and garden care required. There is a makeshift open sided tent beside doors for smokers in wheelchairs - a dismal and drab welcome to D's "home". Sliding doors open to a center hallway with "wards" radiating off the sides. Where is the community gathering?

First thing I think of is the enormous contrast to Canuck Place – homey, comfortable, welcoming: fresh cookies on the counter, tea/coffee and milk/juice are always available. Home like environment with various types of living and family rooms to enjoy. Poppy the dog is usually around. Kids voices and laughter resonate through the house. Kitchen and dining room bring families and professionals together to share meals and stories. Living spaces invite you to stay and be comfortable.

D “lives” in a hospital room in an extended care ward with other residents who except for D, appear to be 45 plus and most 60 plus. The central community room is mostly empty - looks and feels institutional. He has the usual hospital sized room and kit: bed, over bed table, and side table, and they are all CRAMMED full of his belongings. D mostly “lives” in his power wheelchair. Remote control access with one finger allows him to operate his chair and his computer. The computer is D’s only way to access the world: email, phone, online connections and computer gaming. The curtain between his bed and the doorway is closed to keep the room dim. The window looks out over an unkempt grassy area and parking lot. The window drape is mostly closed, so the view is hidden. Lights are off. D is usually playing computer games or sleeping. He finds it very hard to get comfortable in bed, so does not sleep much at night. He dozes through the day in his chair.

D has so few “things”. There are a few family pictures tacked on the wall. He likes to take pictures and run remote control cars. How do you maintain your individuality, have a sense of being your own person when your life is reduced to institutional furniture, a “chair” and computer? What makes you “you” in a place that is not your own?

D is 28 and has lived away from his family home in various institutional settings since he was 20 ish. His immediate family is dispersed across the province and country. (Field Notes, February 14, 2012)

Interviewee two

E is 20. She lives with her parents and younger sisters in a beautiful home on a large property in well-appointed suburban neighbourhood. Large windows showcase the beautiful gardens and pool that surround her home. Being inside in E’s home does not feel far away from the outside. The home was built to accommodate a w/c, so has wide hallways and lots of room to maneuver a chair. Various living areas create comfortable places to be with people or alone. Feels like a very livable, pleasant, caring home and family. Such a contrast from D’s living conditions.

E has silver dot/button on her forehead that allows her to operate her power chair and communication device. She points the button at words, phrases and letters on this device that then appear as typed words on her computer screen. E also “speaks” through vocalizations that her mom is adept at interpreting. She has a monitor on both her shoulders – one for the computer and the other for her w/c.

A power wheelchair with a ventilator and feeding tube equipment overshadows the person in the w/c. E also has a the computer assisted communication device that further creates what feels like a "shield" from connecting with the person inside of all that. I feel intimidated/daunted about connecting with her. I work hard to address my questions to her, and not her mom, maintain eye contact, and find ways to connect with her vocalizations.

E’s mom mostly speaks for her. E vocalizes and her mom interprets, or her mom speaks while “connecting” with E to ensure that what she is saying is what E intends. Their rapport works well - E disagrees and adds to her mom’s comments. (Field Notes, March 15, 2012).

The following table demonstrates the progress and rationale for decisions made through the face-to-face interview process (Creswell).

Table 3

Audit Trail of Decision-Making for Face-to-Face Interviews

Issue	Rationale/Action
Series of short (30-45 minute) face to face interviews	<ul style="list-style-type: none"> • Limited energy levels of participants: easily tired speaking with tracheostomy and ventilation; easily tired and frustrated typing out answers; • Participants not accustomed to long conversations • Opportunity to approach sensitive topics more than once and from varying perspectives
Parent answering questions in face to face interviews	<ul style="list-style-type: none"> • Unaware till arriving at interview that participant only used vocalizations and computer assisted communication; • Preference of participant for mother to “speak for her”

	<ul style="list-style-type: none"> • Participant readily and easily corrected mother’s comments
Using directed versus open-ended questions in face to face interviews	<ul style="list-style-type: none"> • Limited energy levels of participants • Extra energy required speaking with tracheostomy and ventilation • Energy required to type out answers • Participant fatigue • Participants not accustomed to long conversations
Limiting extent of questioning on difficult topics such as end of life	<ul style="list-style-type: none"> • Respecting that participants power and preference to “not go there” • Purpose of questions is interview-based and will not be a long term relationship

Bulletin Board Focus Groups

The purpose of the BBFG was threefold. First, it provided a data collection strategy to reach medically fragile participants, such as young adults with pedLTC. Given the functional, health, and equipment requirements of the participants, convening and conducting in-depth interviews would be expensive and logistically complicated. Second, employing a BBFG with this population provided an opportunity to evaluate the effectiveness and relative merits of an online focus group to collect data from medically fragile populations. Third, the BBFG probed, corroborated, expanded and differentiated the findings of the face-to-face interviews. Table 4 provides the audit trail documentation of the BBFG.

Table 4

Audit Trail of Decision-Making for Bulletin Board Focus Groups

Issue	Rationale and Decision
BBFG instead of OLFG (Online focus group)	<ul style="list-style-type: none"> • BBFG allows time to consider answers and respond to comments of others, so that “introvert” style thinkers and communicators are as equally heard as “extrovert” style thinkers

How many days for the BBFG? Sequential or alternate days?	<ul style="list-style-type: none"> • Facilitates participation as energy level allows • Too many topics for 3 days; hard to keep interest for multiple days; conflicts with attending school • Alternate days may be difficult to remember which days are “on” and “off”
“Soft selling” participation on BBFG to medically fragile young adults with repeat emails	<ul style="list-style-type: none"> • Many young adults do not check email regularly. Use other social media for communication. • Transition coordinator at Canuck Place Children’s Hospice confirmed their interest; slow email response appeared to be a “connection” issue
Securing completed consent forms	<ul style="list-style-type: none"> • Repeat email reminders needed to secure return of consent forms electronically, by fax or mail • If consent forms not returned after 3 requests, participation on BBFG taken as consent
Finding a balance between probing (asking more questions) and overloading	<ul style="list-style-type: none"> • Considered capabilities of each participant gleaned through profile information, and quality and amount of information provided in previous responses • Created a conversational tone and style within parameters that did not provide burdensome amounts to read prior to responding • I am an “older” researcher and they are young adults; therefore, we will likely have different communication styles, especially in online formats
Using text or video	<ul style="list-style-type: none"> • Each day, BBFG was set up with introductions, instructions and questions from me in both text and video formats, providing choice for participants. • I followed the lead of the participants in subsequent responses using either text or video. For short responses, text was preferable. To probe sensitive or subtle topics, video was used to provide more contextual information about what I was asking and be more conversational than directive.

Specialist Informants

Two interviews were conducted with the specialist nurse clinician to provide context to a specific participant’s situation, and to extend my

understanding of palliative neuromuscular conditions. One interview was conducted with each of the remaining informants. Interview questions were specific to the practice specialty of each of these informants and to information gaps arising in the face-to-face interviews. In addition, consultations and conversations with transition committee members from CPCH were also conducted throughout the research process. The practice disciplines represented on this committee and in my consultations included the following specialists in pediatric palliative care: a nurse practitioner, counsellor, physician, teacher, and social worker. These consultations were not recorded and transcribed, but they added context, were treated as field notes supporting the data, and expanded the research process.

Also, as mentioned previously, concurrent to this research, I was managing a project for a provincial professional medical organization to improve transition processes for youth with chronic health conditions. In this capacity, I was collaborating with clinicians in hospital, clinic and community practices working with youth and young adults with complex chronic health conditions, and this enhanced my appreciation and understanding of the scope and intricacies of cross system transitions.

Table 5 highlights the issues arising during interviews requiring specialist informants.

Table 5

Audit Trail of Decision Making to Interview Specialist Informants

Issue	Rationale/Action
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<p>Participant describing efforts to secure staffing, funding, and support for independent living. More information required to understand system requirements and barriers to access.</p>	<ul style="list-style-type: none"> • Housing allocation service provider did not respond to requests for an interview, therefore interviewed pediatric neuromuscular nurse clinician working with participant to secure independent housing.
<p>Face-to-face participants reticent to discuss intimacy and sexuality</p>	<ul style="list-style-type: none"> • Pediatric palliative physician committee member identified discussions about sexuality as a larger “elephant in the room” than death • Identified a post doctoral researcher specializing in sexuality issues for young adults with disabilities to discuss the impact of disabilities and pedLTC on sexuality, and approaches to this discussion on the BBFG
<p>Majority of cognitively competent young adults with pedLTC have a degenerative neuromuscular condition. Further understanding of the breadth of conditions, equipment requirements for management of symptoms, expected disease progression, and preparation for young adulthood and end of life required.</p>	<ul style="list-style-type: none"> • Interviewed expert pediatric neuromuscular nurse clinician.
<p>The literature and participants describe lack of coordinated medical care in adult services. One participant described his exceptional care coordination and support from his primary care provider.</p>	<ul style="list-style-type: none"> • Interviewed this primary care coordinator to understand his approach to this population.

Documents

Documents and other sources were collected from recommendations found in professional, advocacy and interest group, academic, health region, and governmental and non-governmental email circulars, publications, and

conferences. Documents from these sources were reviewed for content related to: 1) young adult development, 2) transition experiences of young adults with pedLTC, 3) recommendations for changes to service provisions, and 4) recommendations for young adult hospice. The small sample size of relevant documents allowed for review of all documents found through these sources. Sampling and review of documents was ongoing and iterative throughout the entire research process and writing of this dissertation. Because young adults with pedLTC are a new population in the early stages of becoming recognized, specific palliative sources are limited, and some literature pertaining to disabilities and complex non-palliative conditions was included. The best source documents are listed in Table 6.

Table 6

Best Source Documents

Sample Document	Most Relevant Content	Source Leading to Document
All About Change (Video) (Merwitzer, 2005)	Young adult with pedLTC experiences	Young adult hospice newsletter
A young adult with pedLTC (Amy-Claire Davies) gives a speech to parliaments The National Council for Palliative Care, (2012, November 12).	Young adult with pedLTC experiences; recommendations for changes to service provisions	Advocacy group newsletter
MIT Young Adult Development Project (Simpson & Kettyle, 2008)	Young adult development: unique strengths and vulnerabilities	Academic conference proceedings
(S)exploring Disability: Intimacies, Sexualities, and Disabilities	Young adult development with disabilities	Young adult hospice newsletter

(Liddiard, 2012)		
Marie Curie Young People and Transition Program www.mariecurietransitionprogramme.wordpress.com	Young adult with pedLTC experiences; recommendations for changes to services; getting on with living a normal life	Professional email newsletter
The Stepp Project The National Council for Palliative Care (2012, November 27).	Young adult with pedLTC prevalence, experiences, recommendations for change	Advocacy group newsletter
Transition: Getting it Right for Young People (Department of Health, 2006)	Experiences of young adults with complex conditions; Recommendations for improvement	Academic paper

Working with Multiple Data Sources

Utilizing different types of data sources within the population of young adults with pedLTC was an intentional research strategy. The face-to-face interviews with the young adults and experts in the field facilitated: 1) an in-depth exploration of the unknown topic, 2) a comparison of key issues identified in the adolescent transition literature with the experiences of these young adults, and 3) further understanding of transitional issues from participants with vastly different experiences of support from their family and community to assist them in navigating adult systems and services. The face-to-face interviews also pinpointed significant and recurring themes, and informed question development for the BBFG.

The BBFG format fulfilled other research aims. First, it facilitated probing the most salient themes identified by the face-to-face participants, and seeking

confirmation, expansion, explanation, paradoxes and alternate points of view. Second, group conversations created interactivity and expansion of ideas that would not have occurred in individual interviews. For example, engaging young adults in a sensitive topic such as how they experience two opposing forces in their life (hoping for “normal” experiences of young adulthood, while experiencing or anticipating a shortened life to live “normal” experiences), improves participation and provides safety in a group. Finally, the BBFG provided data collection opportunities with a medically fragile population that could not be realized with face-to-face interviews or the convening of a focus group. The next paper, Innovations in Research with Medically Fragile Populations, provides a comprehensive and detailed description of the development and implementation of the BBFG. The BBFG can be accessed for review through the following electronic link: <http://login.itracks.com/login>

Username: Observer

Password: exboard9988

Room Number: 43155

This paper will continue with a discussion of analytic methods, followed by the trustworthiness, ethical considerations, and limitations of the study.

Analysis

While described separately here, in case study research, method development, data collection, and data analysis are conducted simultaneously (Creswell, 2007; Yin, 2009). The BBFG discussion immersed me in the data and

revealed early themes and patterns to probe. I had a head start in analysis, and within the BBFG program it was easy to alter questions to follow the participants' ideas and unexpected opportunities; a hallmark feature of case study research (Eisenhardt, 2002).

Unlike other qualitative methods, case study research design does not dictate a specific method of analysis, and Yin (2009) describes analysis as the least developed and most difficult aspect of his research strategy. Therefore, I carefully considered Yin's data analysis strategies such as pattern matching, explanation building, time series analysis, logic models, and cross case synthesis; however, none of these strategies are suitable for a descriptive case study. Pattern matching and explanation building are best suited to explanatory case studies; time series analysis follows changes over time; logic models are best suited to case study evaluations, and cross-case synthesis applies to analysis of multiple cases.

Content Analysis

I chose qualitative content analysis as the analytic method for the following reasons. First, it provides a systematic and objective means for describing the varied experiences of young adults with pedLTC who have transitioned (Elo & Kyngas, 2008). Second, content analysis provides a framework for analyzing the interviews and focus groups, and other data sources such as print materials and observations, thus supporting triangulation (Hsieh & Shannon, 2005). Third, qualitative content analysis aligns with the constructivist

philosophical framework, providing the flexibility to pursue new lines of thinking as data collection and analysis unfold during face-to-face interviews and BBFGs (Baxter & Jack, 2008; Eisenhardt, 2002; Yin, 2009). Finally, content analysis allows for inductive analysis using the propositions as a framework, and deductive analysis of emerging themes and patterns (Elo & Kyngas; Yin).

The goal of content analysis is to provide knowledge and understanding of the phenomena being studied using a subjective interpretation of the text, that includes a systemic classification process of coding to identify themes and patterns (Hsieh & Shannon, 2005). The text data may be in verbal, print or electronic formats and be obtained from narrative responses, surveys, interviews, focus groups, observation or print media such as articles, books, or manuals (Hsieh & Shannon). Therefore, content analysis is well matched to the data collection methods of this case study.

Hsieh & Shannon (2005) have identified three distinct approaches to qualitative content analysis: conventional, directive and summative (Hsieh & Shannon, 2005). Conventional content analysis is used when the intent of the study design is to describe a phenomenon. Directive content analysis is used when prior research or theory exists about a phenomenon that is incomplete or would benefit from further description. Summative content analysis is used to understand the contextual use of certain words or content to discover underlying meanings. This method will not provide the needed depth and scope of the transition experience.

Both conventional and directive content analyses were considered for this study. Conventional content analysis is appropriate when existing theory or literatures are limited. While there is very little literature on the transition of pediatric palliative care patients, there is substantive literature on transitions of pediatric patients to adult health services, and theories about vulnerability, growth and development, and grief and loss. In contrast, directed content analysis provides a more structured framework of analysis through the use of existing theory or prior research to determine the initial coding categories (Potter & Levine-Donnerstein, 1999 in Hsieh & Shannon, 2005). Theoretical frameworks and theory can also provide predictions about the variables or relationships among the variables. For these reasons, both conventional and directed content analysis strategies were used in this study.

Coding

A conceptual foundation of the factors influencing transitions from pediatric to adult services for youth with pedLLC was developed from the theories of growth and development, transitions, grief and loss and vulnerability, and helped to provide initial predictions about relationships among the variables and coding categories. Data display matrices were developed to display the coded data to allow for comparison between and within categories to facilitate further refinement of the emerging categories, and themes (Miles & Huberman, 1994). Matrices were initially set up for each type of data collected (face-to-face interviews and BBFG), and then subsequent matrices were expanded or

developed to encompass all of the data. A constant comparative method among the categories facilitated the identification of patterns, themes, variations and relationships between and among the categories (Miles & Huberman,; Patton, 2002). I singularly reviewed and coded all transcripts. Two committee members each coded one face-to-face transcript.

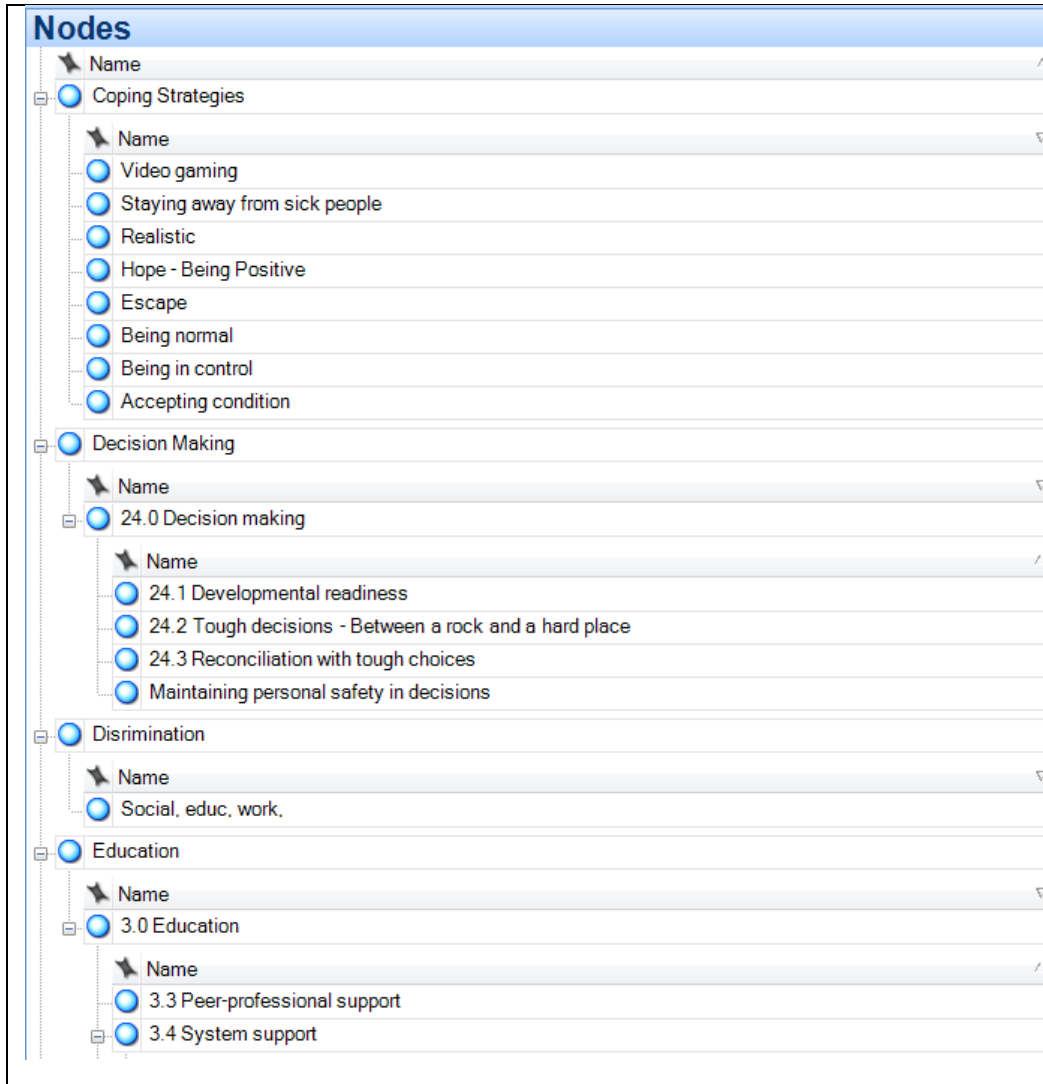
Coding process. Data coding commenced with data collection. In Table 7, the iterative coding process that occurred concurrently through all phases of data collection is described, facilitating a contextually rich analysis of the data.

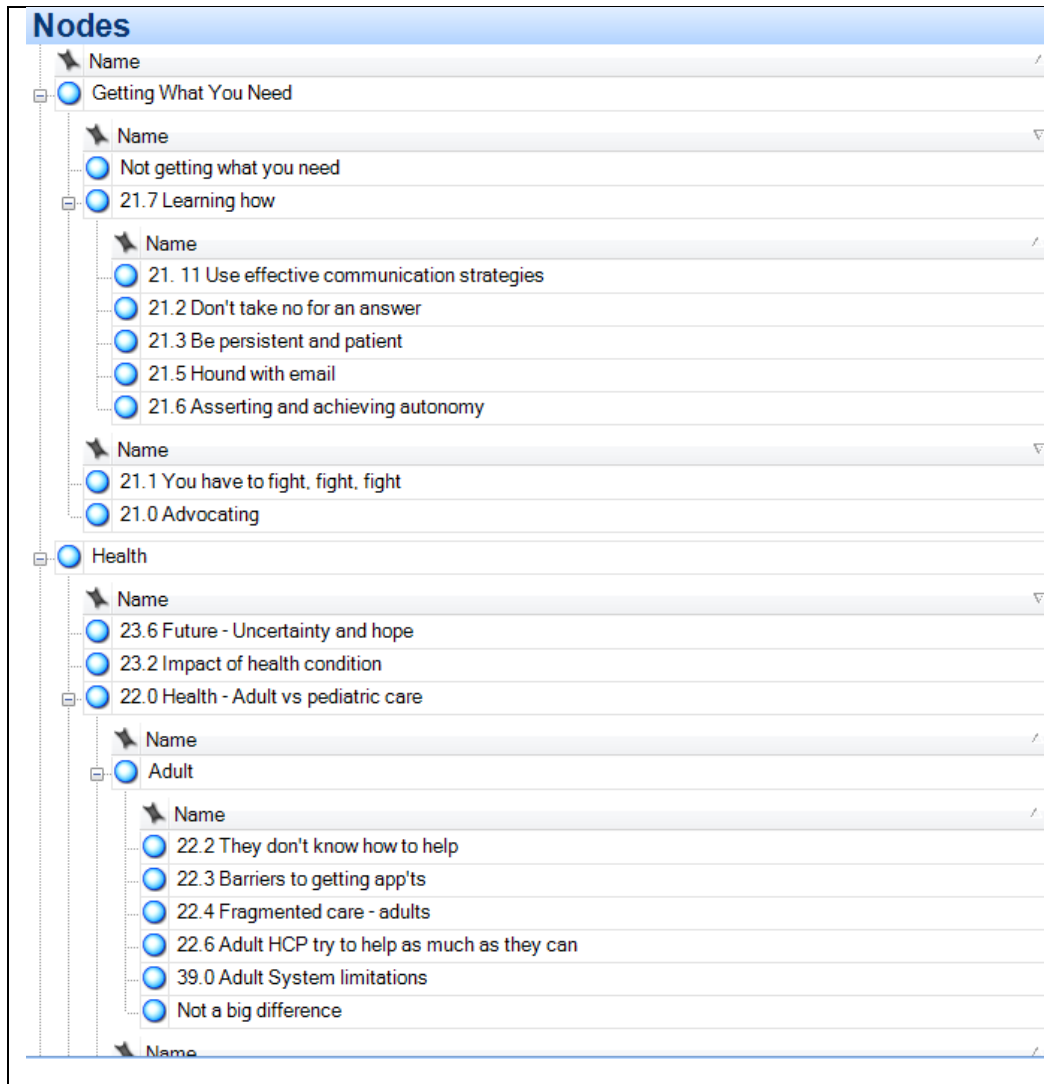
Table 7

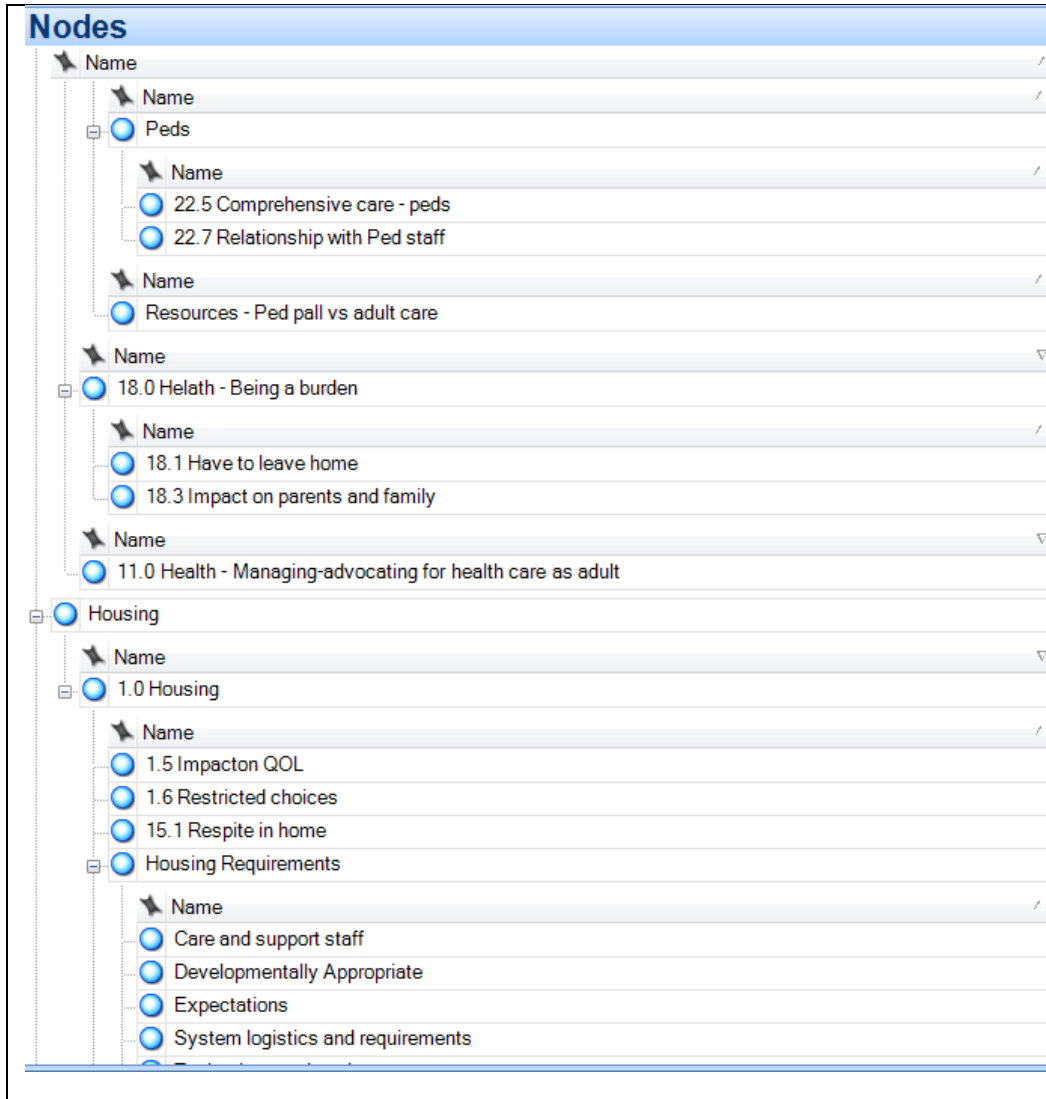
<i>Stages of Data Coding for Face-to-Face interviews and BBFG's</i>	
Stage of Coding and Analysis	Issues and Process
1	First reading and early appraisal <ul style="list-style-type: none"> • Hand coding in margins of face-to-face interview transcripts 1-1, 1-2, 2-1 & 2-2
2	Clustering initial codes (child nodes) into groups and assigning descriptor code (parent node) for group <ul style="list-style-type: none"> • Example: Child nodes: “developmental and medical appropriateness” and “having no where else to go” were clustered under parent node “housing”
3	Establishing credibility and confirmability <ul style="list-style-type: none"> • 2 committee members coded portions of these interviews • Determined agreement of codes (many) and new codes not considered • Performed detailed analysis of developing codes using Excel spread sheet with committee member
4	Software management of data <ul style="list-style-type: none"> • Attended tutorials with NVivo expert, and data management workshops • Careful review and reconsideration of codes → modifications to parent and child nodes that were more contextually representative of the data

5	<p>Analysis using tables and matrices:</p> <ul style="list-style-type: none"> • Categorized all face-to-face codes into 5 groups linked to the major themes of the conceptual framework: Transition, Developmental Stage, Condition Specific and Medical Issues, End of Life and Other. • Compared young adult developmental milestones within and between 2 face to face participants on themes of housing, education, meaningful activities and pursuits, socialization, independence and autonomy, end of life preparation
6	<p>Coding other interview sources:</p> <ul style="list-style-type: none"> • Focused on expanding contextual dimensions of the following parent nodes: medical/condition specific, housing (especially barriers to independent living), end of life preparation, disability and sexuality, and advocacy
7	<p>Coding of BBFG's:</p> <ul style="list-style-type: none"> • Perspective of coding changed because participants in BBFG expressed fewer limitations and more opportunities <ul style="list-style-type: none"> ▪ For example, socialization child node themes such as “not fitting in” were expanded to include “forgetting that I have a disability” • Further expansion and refinement of parent and child nodes
8	<p>Further analysis using tables and matrices to compare across groups of participants (face-to-face and BBFG) and within BBFG groups</p>

Development of codes. As described in Table 5, codes were developed and modified through a continuous iterative process that included free coding of initial transcripts; clustering related codes into parent nodes; defining and delineating the scope and dimensions of the nodes; and modifying parent node names and clustering child nodes as new data sources provided new understanding and illumination. Unlike NVivo 8TM and 10, NVivo 9TM does not support creating and exporting the list of parent and child nodes. Figure 3 provides screen shot examples of parent and child nodes developed and refined for this study, but is not the complete list.







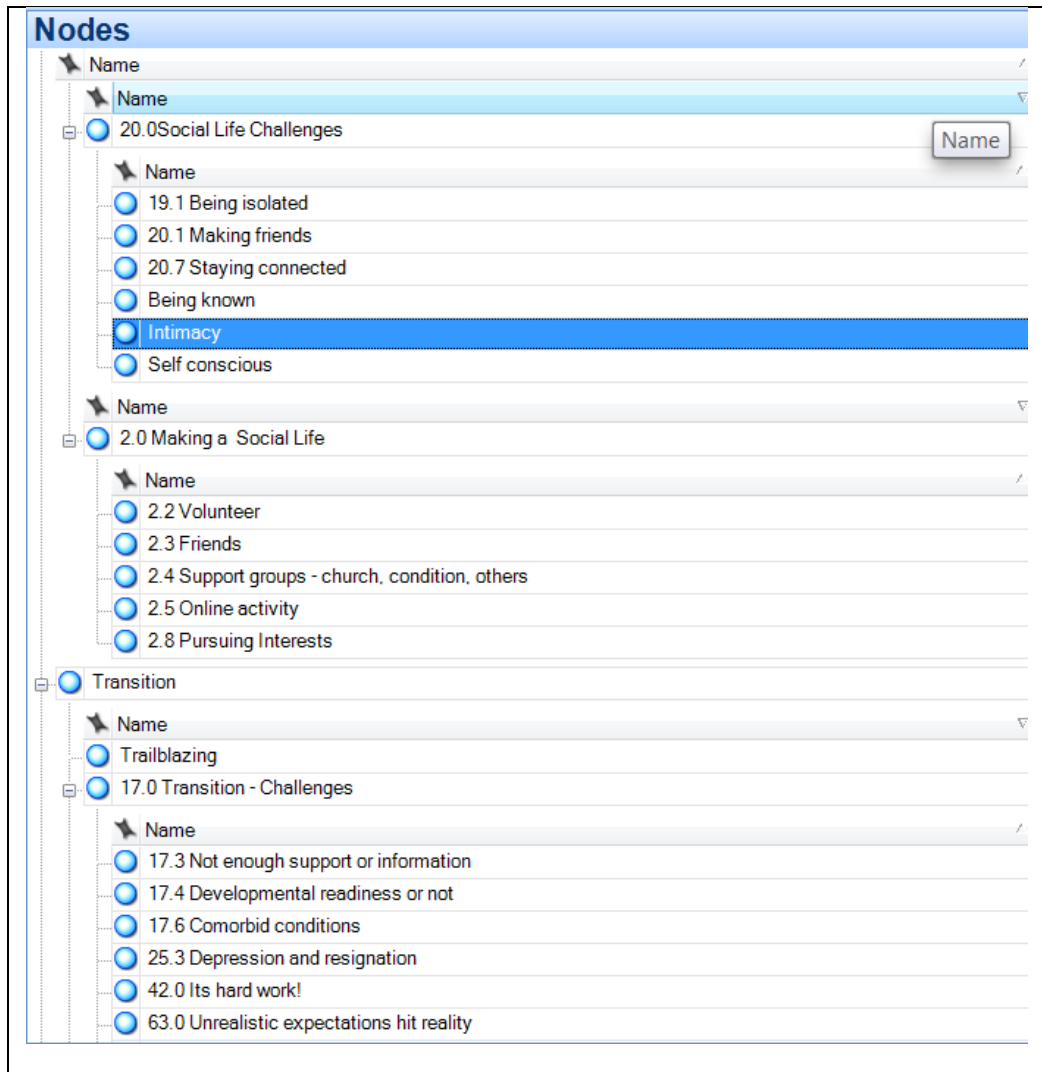


Figure 3. Parent and Child Nodes using NVivo 9™

Analysis using matrices and tables. Data display matrices served several purposes. First, the dimensions (breadth, depth, similarities and differences) within parent nodes were examined and described (S. Jack, personal communication, June 21, 2012; Miles & Huberman, 1994). Second, matrices were used to cluster related parent nodes within larger categories and then compare and contrast the child nodes that characterized these categories (S. Jack, personal communication; Miles & Huberman). For example, during early analysis of the

face-to-face interviews, categories describing “difficult transition experiences” and “transformational experiences” appeared to be both juxtaposed and connected. A matrix was constructed to: 1) display the parent nodes that align within each of the larger categories (difficult transition experiences and transformational experiences), 2) facilitate comparing and contrasting the dimensions of the child nodes providing different meaning to the experiences, and 3) provide new exemplars that refine and expand the emerging categories and themes (Miles & Huberman). In Table 8, the use of a data display matrix demonstrates differing participant experience outcomes within the same parent nodes. Reading across the table demonstrates the contextual differences of child nodes within the same parent node, and the other additive factors that transformed their experiences. In this example, different child nodes describe both difficult and transformational experiences within the same parent node, and additional nodes in the transformational experiences category provide new exemplars that refine and offer further understanding of their experiences. This data display matrix is a portion of a larger matrix including all of the relevant nodes.

Table 8

Demonstrating Matrix Use to Compare and Contrast Parent and Child Nodes Within and Across Categories.

Category: Difficult Transition Experiences		Category: Transformational Experiences	
Nodes	Exemplars	Nodes	Exemplars
Housing: restricted choices	Nowhere to go except extended care/nursing home	Housing: securing independent living arrangement; Getting What You Need: not taking	“You have to fight, fight, fight!”; Don’t take no for an answer; Family

		“no” for an answer	support when things get hard
Social: Social Life Challenges	Not fitting in, challenges staying connected; being housebound; losing communication abilities; wishing you could be with your friends, and be more like your friends; not being able to have a boyfriend because “there’s nobody out there.”	Social: being known; online activity; volunteer; pursuing interests.	Autobiographical writing; testimony; making cards to raise money for charity; writing a book; “loving being the girl in the chair”; power wheel chair sports
Health: Being a burden	Have to leave home before developmentally ready; unable to be left alone; family limitations for holiday; rely on for care	Health: Taking risks Decision Making: Personal safety	Staying home alone without attendant, leaving extended care facility without attendant (for ventilator and suctioning support). “I am an adult and can make my own decisions for myself.”
Finding Meaning: Frustrations	No work or volunteer opportunities due to condition, equipment (e.g. power w/c) getting in the way; volunteering at	Finding Meaning: Pursuing meaningful activities; Transition: Trailblazing; It’s Hard Work	Attending college; painting and card making; play power soccer; unable to cook, but directs others to cook; photography, model airplane

	cancer society and then being diagnosed with cancer; education programs not flexible to accommodate		flying
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Third, in the next stage of analysis, matrices provided a framework to interpret the data within specific propositions (Miles & Huberman, 1994). The following matrix (Table 9) demonstrates the alignment of specific nodes and their exemplars with one of the study’s propositions: “Continuity of appropriate medical and support services did not follow interventions to increase life expectancy” (Hamdani et al., 2011). Interpretation of the data revealed both predicted theories and themes from conceptual framework, and a new emerging theory (Miles & Huberman). In the following table, the proposition encompasses the relevant nodes and their key exemplars, predicted coding theories and themes from the conceptual framework (growth and development, transition, loss and vulnerability) and a new theory, coping and appraisal (Lazarus & Folkman, 1984). This matrix demonstrates how the analysis process advanced and refined the body of theories from the pre-study conceptual framework.

Table 9

Matrix Demonstrating Code, Theme, and Theory Development Related to Study Proposition

Proposition: Development of comparable adult medical and support services did not follow interventions to increase life expectancy (Hamdani et al., 2011).				
Child Nodes	Parent	Key Exemplars	Interpretation	Themes and

	Node			Theory
Manage-advocate for adult health care		We have a book that we wrote minute-by-minute, blow by blow, E's day. . . to account for how many hours of care E needs in a day in order to put our case for how many hours we were going to get because they were going to cut you, they'll cut you down	Successful access to services achieved through empowerment: (personal agency and strategies, anticipatory guidance, perseverance)	Empowerment; Coping; Transition barriers
You have to fight, fight, fight!		Pretty much its fight for what you want, don't take no for an answer, and just keep trying until you get what you want, or get what you need. You've just got to keep fighting.	1) Transformation: Shifting powerlessness into success; 2) Frustration with systems provides meaningful involvement; 3) "the fight" is an outlet for frustration	Empowerment; Accessibility; Vulnerability; Transition barriers; Resilience
Learning how to advocate	Getting what you need: advocacy skills	1. Research what services are available to you and how they work/how to start receiving them 2. Talk to other people in similar situations that you are in! 3.	Successful access to services achieved through empowerment: (personal agency and strategies, anticipatory guidance, perseverance)	Empowerment; Transformation ; Accessibility; Coping; Transition barriers; Resilience

		<p>Be persistent 4. Remember that these services are for you, that's why they are in place 5. Use personal appeals to pull the heartstrings, to emphasize your needs by services I am referring to things like grants and funding that you are entitled to, even if it seems otherwise and tricky to get on to or to find information about.</p>		
<p>Using effective communication strategies</p>		<p>For me I did most of my negotiations by myself, but I had people such as doctors and OT's and physios to support me, so it would be my idea, but like if we were ever confronted such as at <i>university</i> when we wanted an accessible washroom, I was the one that talked about the idea and. . . the</p>	<p>Successful access to services achieved through empowerment: (personal strategies, anticipatory guidance, perseverance)</p>	<p>Empowerment; Accessibility; Coping; Resilience</p>

		advisors couldn't turn me down.		
Not taking no for an answer		I had to fight with a team of OT's and social workers and my physio for a ceiling lift in the washroom and also a bed which I can lie down. And even though the disability advisor at first didn't want to listen to me after awhile and she saw that I really, really wanted to go to <i>university</i> she made it possible. So even if someone tells you that they can't do anything just keep on trying and eventually you'll be able to get what you want.	Successful access to services achieved through empowerment: (personal agency, strategies, environmental supports, anticipatory guidance, perseverance)	Empowerment; Transformation ; Accessibility; Coping; Transition barriers; Resilience
Be persistent and patient		In my first year of university I was met with the challenge of working with the Centre for Students with Disabilities (CSD) at <i>university</i> to	Successful access to services achieved through empowerment: (personal strategies, anticipatory guidance,	Empowerment; Transformation ; Accessibility; Coping; Transition barriers; Resilience

		<p>ensure that my needs where met in an adequate manner. I wanted to create an environment for myself where I could actively compete with other students. Due to the effects of my disability, I require the support of a personal care attendant to assist me in setting up study materials and meeting my personal care needs. Initially, I found that the university was unprepared to assist me because my “needs are too intensive,” and I felt that what I had to offer as a student was not valued. I continued to pursue my studies while researching my rights as a student and how the CSD could assist me in</p>	<p>perseverance)</p>	
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		<p>maximizing my educational pursuits. I decided pay my attendants' wages out of my own pocket while I worked with the CSD to seek outside funding, and I was eventually reimbursed. Throughout this process I discovered that I can find greater success when collaborating directly with the institutions advocating for students with disabilities, rather than passively waiting for my needs to be met.</p>		
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In summary, constant and comparative coding facilitated the identification of patterns, themes, variations and relationships among nodes and between categories to examine the participants' experiences in relation to the study's proposition and conceptual framework, and illuminate further expansions (Miles & Huberman, 1994).

Methodological Rigor: Trustworthiness

Triangulation of data is the hallmark of methodological rigor in qualitative research (Eisenhardt, 2002; Baxter & Jack, 2008; Yin, 2009); thus, references are peppered throughout this methodological review. Briefly, the multiple data sources in this study include two different types of interviews with young adults; interviews with health and social service providers; review of documents, such as practice reviews, recommendations and policies; and my own observations as a researcher and professional in the field (Yin, 2009). Further, the components of Lincoln and Guba's (1985) criteria for trustworthiness—credibility, dependability, and confirmability—are also widely accepted standards for the evaluation of qualitative studies and are examined below. Transferability, the fourth criteria of trustworthiness, will be considered in the context of a small sample of young adults with pedLTC within their specific contexts, and studied in depth (Miles & Huberman, 1994).

Credibility

Credibility is the believability of the findings (Creswell, 2007) and the level of confidence in the findings verified through prolonged engagement with the participants and data, persistent observations, triangulation, memo writing, member checking, and searching for disconfirming evidence (Creswell; Lincoln & Guba, 1985). Multiple data sources have been described. Triangulation of the multiple data sources was achieved through 1) prolonged engagement with participants in face-to-face interviews and BFGs, documents and data, 2) persistent observations made possible in multiple face-to-face interviews with the

same participants, and inherent in conducting a BBFG over five days, 3) participants reading and responding to the study results (member checking) and, 4) the constant searching for disconfirming evidence in the analysis process (Baxter & Jack, 2008; Eisenhardt, 2002; Yin, 2009). The following comments (Table 10) from participants about the results presented in the next two chapters demonstrates the credibility of the findings:

Table 10

Participant Feedback on Study Results

I think it really embodies all of our feelings, goals, and expectations! I especially like the part about the need of having a Young Adults' hospice facility.

Just some other terms I really liked were the comments on "continually constructing a new normal" and the adapt-adjust-transform because it such an accurate idea of us as young adults with life threatening illness'. Always the chameleon, always adapting.

I particularly liked your descriptions of the transition from child to adult as an "abyss" and "like falling off a cliff".... I think those are strong & accurate metaphors for what we described in group. You mention vulnerabilities and resiliency in the very beginning and I really liked how the words (vulnerability and resiliency) were kind of weaved through out the entire paper as it does encapsulate the theme and tie it all together.

All of our topics of discussion (vocation, socializing, intimacy, optimism, fighting for your future, etc. etc.) you've summarized were perfectly interpreted and genuinely represent what was said in my group and also what I had participated.

Given the relative newness of BBFG in nursing research, specific consideration of the credibility of this data collection method is warranted. BBFGs can strengthen credibility in three other important ways. First, high response and retention rates demonstrate that the participants were authentically interested in the study and engaged over a long period of time, thus positively

influencing the credibility of the data (Im & Chee, 2006). Second, the immediate generation of transcripts directly from the participants' responses, including every word, textual descriptors (line breaks, font size and colour, punctuation, and emoticons) and emphasis, ensures a verbatim record without bias or interpretation (Kenny, 2005). Third, the BBFG creates a constant opportunity for member checking. As participants post responses to both the researcher and other participants, they are able to comment on, add to, or provide confirming or disconfirming information to every comment posted.

Dependability

Dependability is the “quality control” measure in qualitative research. In this study, data collection and analysis followed a detailed research process outlined in a proposal to academic supervisors and two university ethics boards (Appendix F). Memos were written after every face-to-face interview, and shared with two academic advisors to expand my perspectives and hunches about the research. Detailed audit trails of the development of the face-to-face and BBFG interview guides, and implementation of these research methods, have already been described (Miles & Huberman, 1994).

Potential data dependability risks specific to this study may have included declining cognitive and psychosocial abilities due to disease progression, forgetting or misplacing the BBFG log-in information, and internet instability dependability (Fox, Morris & Rumsey, 2007; Im & Chee, 2006). However, in this study, none of these potential problems arose.

Confirmability

Confirmability is determined by the degree that two or more independent reviewers agree on the data's meaning and relevance (Im & Chee, 2006).

Confirmability can be compromised when the researcher assumes multiple roles, such as interviewer, BBFG moderator and observer, and data analyst (Franklin & Lowry, 2001). Training in family therapy assisted me in this research process to restrain my clinical and research assumptions and focus on following the participants' stories. Conversations with the BBFG group about the meaning of their online activity, and being dependent enabling their independence, are examples where following the participants' leads overrode my assumptions. Further, careful memo writing and audit trail documentation addressed my queries about the overlap of these roles in the research process (Franklin & Lowry).

Prior to starting the BBFG, the use of abbreviations, emoticons and symbol groupings (e.g. smileys) to convey added meaning to the text were considered as both potential threats to confirmability (East, O'Brien, Jackson & Peters, 2008; Fox et al., 2007; Kralik, Price, Warren & Koch, 2006; O'Connor & Madge, 2003) and benefits to enrich the quality of text-only data. In this BBFG, emoticons, capital letters, punctuation, varied font colors, sizes and styles, and other creative stylistics were encouraged to provide more contextual information. The meanings of these additions to this BBFG were very clear; if they had not been, further questions to the specific participants would provide clarification (East et al.; Fox et al.; Kralik et al.). As mentioned previously, in other

circumstances in the BBFG discussions, follow-through on conversation leads was not always achieved, but did not compromise understanding.

Ethical Considerations

To uphold the ethical standards of qualitative research and ensure the consent and confidentiality of the participants, the face-to-face interviews and BBFG were prefaced with a discussion (in person or on-line) of the following issues: the purpose of collecting the information, who would use the information and how it would be used, what kinds of questions they would be asked, how the information provided would be recorded, transcribed, stored and maintained confidential, and potential risks and benefits of participating in the study (Creswell, 2007; Patton, 2002). In addition, participants received recruitment and consent letters that contained this same information. Face-to-face participants signed a permission request to record and transcribe their interviews (Creswell; Patton). Forms returned via email with typed signatures served as the informed consent for the BBFG participants. One participant did not return a signed consent form. After three requests for the form, participation on the BBFG was taken as participant consent.

Participation in face-to-face interviews and the BBFG was confidential and voluntary. Face-to-face participants were asked at the end of each interview if they were willing to continue, and were provided with a phone number and email address to inform the researcher if they wished to decline further interviews.

Online focus group participants could withdraw at anytime by not participating in the online discussion, or notifying me via email.

While no significant unwanted effects were expected, contact information (telephone, online, or in person) for access to a teen counsellor known to the youths was provided prior to the start of the face-to-face interviews and the BBFG. The topics probed in these interviews may elicit memories, or hopes of dreams never fulfilled, or reflections on decisions made long ago (Patton, 2002). However, the youth who have participated in such discussions at CPCH teen camps have expressed appreciation for the opportunity to share their experiences with their peers (S. Poitras, personal communication, October 11, 2012). I monitored the nature, tone and content of comments posted on the BBFG for signs of distress, and the reactions of other participants to the comments.

Ethical Issues for Face-to-Face Interviews

To protect the confidentiality and privacy of the participants, code numbers were used to identify all personal information in documents, which were kept in a locked cabinet or a password-protected computer. Participants are not identified by name in any reports of the completed study; however, participants may be identifiable through the stories they tell. All of the files with identifying information will be destroyed 10 years after the study is complete.

A professional transcriptionist was hired to transcribe the face-to-face interviews to ensure completeness, accuracy and confidentiality, and the electronic files will be destroyed when data analysis is complete. All the raw and

transcribed data masked names and identifying characteristics. The data files were managed electronically on a secure network equipped with a firewall. Hard copy back-ups were secured in a locked cabinet for the duration of the study and will be destroyed when no longer required for verification or publication purposes (Creswell, 2007).

Ethical Issues for Bulletin Board Focus Groups

By virtue of participating in a BBFG, the participants were sharing their stories with each other. They were encouraged to upload an avatar or picture to enhance group dynamics in an online format. Some participants recognized each other from their stays at Canuck Place Children’s Hospice. On the preliminary discussion board, the participants were advised to respect the privacy of their fellow participants by not repeating or sharing the stories they read on the BBFG. iTracks™ disables downloading of webcam video clips with a “right click”; however, as with any online activity, screen shots cannot be prevented.

Confidentiality of the data collected through the online focus group was protected through the comprehensive security measures of the “iTracks™” software system, which includes the hardware, software, data security and storage. “iTracks™” is a Canadian company that services more than 2500 online groups every year. The online data collected is only stored and accessed in Canada. The online focus group is only accessible with a unique code and password that is provided by iTracks™, and only the investigator and participants had access.

Conclusion

This paper has provided a comprehensive overview of the qualitative case study methods undertaken in this study, and described the procedures and rationale behind decisions made in the research process in order to ensure the rigor and trustworthiness of the results. Further description of this study's methodology continues in the next chapter. Chapter 3 is a manuscript in press that describes a significant component of the methods work for this study: the background, development and implementation of an innovative data collection strategy for medically fragile populations. Some overlap and repetition between these two papers is unavoidable, to ensure the reader has a complete description of the methods employed to conduct this study.

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INTRODUCTION TO CHAPTER 3

This manuscript is both a continuation of Chapter 2 Methods, and an in depth description of the rationale, development, and description of an innovative data collection method for medically fragile participants. At the time of writing, this manuscript is awaiting editorial review with *The Qualitative Report*, and follows that journal's formatting style. Portions of this paper were presented in June 2013 at the *Advances in Qualitative Methods Conference* held in Edmonton, Alberta.

Electronic access to Bulletin Board Focus Group (BBFG) can be accessed for review through the following electronic link: <http://login.itracks.com/login>

Username: Observer

Password: exboard9988

Room Number: 43155

CHAPTER 3

INNOVATIONS IN RESEARCH WITH MEDICALLY FRAGILE

POPULATIONS:

A CASE STUDY

New Population of Medically Fragile Patients

Advances in pediatric care have not led to an increase in appropriate support services for young adults with pediatric life threatening conditions (pedLTC) that live past 18 years of age (Doug et al., 2011; Hamdani, Jetha & Norman, 2011; Stewart, Stavness, King, Antle & Law, 2006). A new group of medically fragile young adults with conditions such as Duchennes Muscular Dystrophy (DMD) is graduating from pediatric palliative care programs with limited expectations to live beyond early adulthood, and there are no comparable adult health and social services to support their complex needs. Furthermore, the philosophical differences between pediatric and adult palliative care exclude this population from adult palliative care services. Whereas pediatric palliative care is lifelong until death, adult palliative care focuses on the last weeks and months of life (Fraser, et al., 2012). Very little research has focused on the experiences of young adults who have transitioned (Fletcher-Johnson, Marshall & Straatman, 2011), and there is no evidence in the literature of transition experiences of youth “aging out” of pediatric palliative care (Doug et al.).

The adult healthcare system is poorly prepared to deal with this new vulnerable population at risk of poor health outcomes (Brown, Katherine, Allen, Quach, Chiu & Bialystok, 2010; Prestidge, Roman, Djurdjev & Matsuda-Abenedini, 2012; Meleis, Sawyer, Im, Hilfinger & Schumacher, 2000). These young adults transition out of a comprehensive and supportive pediatric palliative care program into an environment with no specific care delivery system in either palliative or

chronic care services. This gap leaves them striving to achieve young adult developmental milestones within a compressed time frame, while navigating new health, education and social systems that provide fewer supports (Prestidge et al., Meliess et al., 2000; Rogers, 1997). Furthermore, they might be contending with the enormous psychological issues embedded in life limiting conditions, in addition to the impact of their condition on themselves and their family (Young et al., 2009).

The best examples of this health care transition are confined to transfers between pediatric and adult subspecialty clinics that are condition specific, such as diabetes, cystic fibrosis and congenital heart disease (Doug et al., 2011; Kennedy & Sawyer, 2008), and on specific skills and knowledge for successful transition at the individual or health provider level (Hamdani et al., 2011). To date, no research has focused on the experiences of young adults with pedLTC who have transitioned from comprehensive pediatric palliative care programs to adult services. Outside of the United Kingdom, young adult hospice programs and comprehensive adult clinical programs are absent.

My research goal was to understand the experiences and perceptions of individuals within this population, however, there are significant challenges using traditional methods of qualitative data collection, such as interviews or focus groups. Accessing this population is difficult because of the complexity of the individuals' conditions, their limited mobility to travel and commit to specific interview times, and divergent communication abilities due to weakness, that lead

to the use of computer mediated voice recognition, typing or speech. Most of these individuals require 24-hour attendant care, and their independent function can be reduced to minimal movement of their index finger on a joystick to control power wheel chairs, computers and phones. Because of the sensitivity of the topics discussed with this group and the complexities of their condition, I found it vital to select research strategies that would provide meaningful data while remaining mindful of participant communication limitations, and ethical issues that could arise with new research modalities. Because loneliness and isolation are common experiences for those without work or school opportunities, this research method provided the added benefit of a social connection with their peers.

To address these specific methodological challenges, I undertook a descriptive case study (Yin, 2009), using a Bulletin Board Focus Group (BBFG), with the purpose of examining the complexity of factors affecting the transition experiences of young adults with pedLTC to adult services. Bulletin board focus groups (BBFG) are an asynchronous modification of an online focus group. The BBFG conversation is carried on over several days instead of in a single session, allowing participants to log in and out of the discussion when it is convenient for them. Unlike many qualitative techniques that favour those with quick responses (typically extroverts), the asynchronous BBFG creates advantages for those who are less spontaneous. Some question types require more extended thinking, and results can be improved when participants can take time to reflect before answering (Abbott, 2011). This proved especially true of questions relevant to this

study, such as thoughts about the future, intimacy, and end of life preparation and preferences.

In this paper, I describe how the innovative BBFG methodology facilitated access to this medically fragile population, provided a rich qualitative understanding of their experiences and offered benefits to the participants. This research involved medically fragile young adults with pedLTC, not expected to live beyond their first decade of adulthood (19 -29 years). I selected a purposeful sample of English speaking young adults with pedLTC, such as DMD, or Spinal Muscle Atrophy (SMA), who were cognitively capable to participate in a BBFG, from the graduates of a children's palliative care program in western Canada. Two groups of four young adults participated in this five-day BBFG supported by iTracks™, a Canadian software company. The primary investigator is a nurse and clinical counsellor with more than 10 years experience with youth and families managing chronic and palliative conditions, and this study was reviewed and approved by two university ethical review boards. An audit trail of the research process was maintained.

Overview and Rationale of BBFGs with Medically Fragile Populations

Overview

The following section describes the research opportunities that online focus groups offer that cannot be realized with face-to-face interviews or focus groups with medically fragile populations. First, an online format connects individuals geographically disparate from each other (Cantrell & Lupinacci, 2007;

East, O'Brien, Jackson & Peters, 2008; Fox, Morris & Rumsey, 2007; Kralik, Price, Warren & Koch, 2006; Stewart & Williams, 2005) and those requiring high levels of support, such as attendants, suitable and timely transportation, and well functioning equipment, to attend a face-to-face meeting. Second, within this group of young adults, many are losing the ability to type (even with technological aids) or to speak clearly. The BBFG format provides the option to choose either text or webcam methods to participate.

Third, BBFGs reduce inhibitions, so that participants report more freedom to respond candidly and participate in topics that might be considered embarrassing (Campbell et al., 2002; East et al., 2008; O'Connor & Madge, 2003). This is an important consideration for discussion of new developmental issues for these young adults, such as their experiences and desires for intimacy. For many, their appointments with health care professionals have always been (and can still be) in the company of their parents, and they likely have not been provided with the privacy to discuss some of their questions and concerns. Further, teens and young adults are empowered in online formats to be more assertive and confident in their interactions with the adult facilitator than in face-to-face groups. Their confidence and fluency in online social interactions, absence of time constraints, anonymity, and fewer inhibiting social cues create an enhanced sense of control and a willingness to explore sensitive topics (Campbell et al.; East et al.; Fox et al., 2007; O'Connor & Madge).

Rationale

The suitability of the asynchronous BBFG modality for this medically fragile population and the benefits for the researcher are described here. First, it is important with this population for participants to be able to respond in their own time over several days if unexpected personal or medical situations arise, to take more time to reflect on questions, and to “catch up” later. For example, some participants may easily fatigue, or have limited attention spans, or learning disabilities. In the BBFG, questions are evenly paced over several days so that participants can log on and off as often as needed to complete the questions each day, or over several days. The BBFG format also provides the opportunity to balance easy to answer questions (just needing a check mark) with open-ended questions. Online research with this age group using a synchronous discussion has produced frustrating results, confirming the benefits of the asynchronous format (Levine, Madsen, Wright, Barar, Santelli & Bull, 2011)

Second, the BBFG provides potential for the researcher to interact with young adult participants via their preferred communication method (text or webcam), creating an inviting and engaging conversation. Customized responses, daily welcome and wrap up messages, mood and content setting visuals and video uploads can be used to stimulate participants’ imagination and provide various modalities to engage their interest. Participants can upload pictures and images that depict their feelings in addition to their text and webcam responses (Barnes, 2012; iTracks, 2012; Van Patten, 2011).

Third, offline research activities such as projective exercises, ethnographic reflections and video projects can provide depth and dimension unavailable in live or online groups (Barnes, 2012). For example, in a BBFG, a question such as “What does the word ‘health’ mean to you?” can be expanded to: “Please find an image that best reflects what health means to you.” From the images posted, the moderator can stimulate conversation with and between participants to compare the meaning of their image with others. Ethnographic opportunities are opened up, with participants uploading videos and images they have of themselves, so that the researcher can “see” descriptions in addition to text based responses.

Fourth, for groups where fatigue and physical limitations are not a limitation, homework assignments such as recording journals, making physical or online collages, finding or creating pictures and videos all become possible data collection methods, enhancing traditional talk or text based formats (Barnes, 2012).

Finally, the BBFG formats can strengthen credibility of the research through 1) high response and retention rates, indicating that participants were interested and engaged over a long period of time (Franklin & Lowry, 2001; Im & Chee, 2006), 2) the immediate generation of transcripts directly from the participants’ responses, including every word and textual descriptor (Kenny, 2005), and 3) a constant member checking, as participants post responses to both the researcher and other participants.

Development of the BBFG

Phase One: Question Guide Development

To develop questions for the bulletin board focus group, I utilized a content analysis of in-depth face-to-face interviews with young adults with pedLTC, with specialists in pedLTC and disabilities, and a review of relevant literature. It was vital to choose the most salient themes, issues requiring further investigation, and topics most amenable to an online format for question development.

I utilized specific writing techniques that have proved successful and amenable in online formats (Barnes, 2012; iTracksTM, 2012; Van Patten, 2011) to create these questions. In face-to-face group or individual interviews, the moderator's question guide serves as a prompt for the discussion and allows adaptations for participants' responses and body language. In the online asynchronous discussion, the moderator guide is an actual script with a deliberate approach to structure and clarity (iTracksTM; Van Patten). Whereas the BBFG format requires pre-meditated structure, my role was to create a forum that appeared inviting, informal and engaging to the participants (Barnes; Van Patten). For example, I worded the following introductory segment to encourage participant to post divergent points of view in all writing styles, providing contextual information in addition to the text.

Different viewpoints and feedback on the topics are encouraged. And, if you see someone else talking about an experience you share or feel the same way about, add your thoughts too!

Don't worry about spelling, typos or grammar. I'm shur we'll be abul 2 figure out what ur saying. Feel

free to use emoticons :-) coloured font, CAPITAL LETTERS, videos and pictures . . . be *cReative!*

Figure 3. Bulletin Board Focus Group Instructions to Participants

Utilizing the BBFG required specific consideration of the ordering and flow of the questions. To avoid frustrating redundancy and repetition, I anticipated responses to each question and grouped naturally emerging topics next to each other, creating cohesive groupings of questions to create natural breaking points between the daily discussions (iTracks™, 2012). Lacking the ability to improvise the wording of questions in response to previous participant comments, or to add more context and content *ad-hoc*, I found clear and complete online questions necessary to ensure that participants knew how to answer the questions being asked, and to enhance the quality and quantity of their responses. Whereas structure and clarity are the backbone of the BBFG guide, personality and flow are essential for success (iTracks™; Van Patten, 2011).

To ensure that the discussion was a rich and engaging dialogue among group participants, I employed diverse question and response options (Barnes, 2012; Van Patten, 2011). A range of question types stemmed from viewing a video, reading a short vignette or a pictorial representation of the issues to build rapport, match varied interests, and open up possibilities for a range of answers (Teachman & Gibson, 2012). I anticipated that new comments and video responses would create a fresh and responsive virtual discussion among participants and moderator, and encouraged participants to provide more understanding and contextual information than text or webcam responses allowed,

through questions that prompted them to upload images, videos or songs (Barnes, 2012). For example, in this BBFG I asked participants:

When you think about the future, what kinds of thoughts and feelings come to mind for you? If you can, please upload an image or picture that represents your feelings and thoughts about the future. Tell us what your picture or image means to you about the future.

Unlike a face-to-face focus group, where every participant does not answer every question, this BBFG required a response to each question before a participant could move on to the next. If participants preferred not to answer a question, they were prompted to type, “will come back” and then proceed with the next questions. If they were unsure of how to answer a question, they could review what others had said.

BBFG question development requires advance decision-making about the type of question required for participant responses. The following list describes the parameters for question and response types:

- Type of question: Open text, forced choice (yes, no, maybe, don't know), rating scale and multiple choice style questions with the option of one or multiple answers
- Influenced or uninfluenced answers: Determines whether participants can see others' responses before their own, or if must they write a response first before viewing the other participants' responses
- Sequential or non-sequential: Determines whether questions must be answered in sequence, or if participants can skip around

- Time of launch of questions: When will each question or set of questions “go live”?
- Group/segmenting: Provides the opportunity to pose certain questions to specific respondents. Selected participants will see these questions, whereas others will not. Useful for segmenting questions between groups such as male/female, under/over certain ages, with disease/without disease (iTracks, 2012).

Finally, it was important to collect some data regarding topics and themes that were not amenable to an online focus group discussion (for example, questions about with who and where they live, support required, current work, school or volunteer activities and accessibility). Instead of asking these questions in an online discussion, I presented them a day ahead of the “live” discussion, within the Profiles Section of the online forum. Participants could view each other’s responses here if they were interested.

Phase Two: Testing

I developed several iterative drafts of the moderator guide through feedback from expert practitioners in pediatric palliative care, transition and neuromuscular diseases, a young adult with DMD, and academic supervisory committee members. Screen by screen review of the BBFG was tested and reviewed in cooperation with the host software company, ensuring compatibility of the requirements of participants for each screen with the software functionality. Examples of compatibility functionality testing included: video clips, forced and

not forced answer screens, combined open and closed answer screens, optional response screens, and functionality and ease in uploading videos and images into the BBFG.

Managing Ethical Issues

Young adults who met eligibility criteria were contacted via email and invited to participate in the online focus group. We exchanged e-copy consents via email, with returned typed signatures serving as the participants' informed consent.

BBFG members were advised that although every precaution was taken to ensure confidentiality, it could not be guaranteed. Prior to entering the BBFG, participants read and signed a Terms of Service of Agreement with the host software company (iTracks). In addition to abiding by the company's rules of conduct, participants were advised to respect the privacy of their fellow participants by not repeating or sharing the information they read on the BBFG. Confidentiality of answers recorded via webcam was maintained through disabling the downloading and copying functions. As with any online activity, screen shots could not be prevented. Confidentiality of the data collected through the online focus group was protected through the comprehensive security measures of the "iTracksTM" software system, which includes the hardware, software, data security and storage.

I provided each youth with contact information (telephone, online, or in person) for access to a teen counsellor already known to them. Additionally, I

closely monitored the nature, tone and content of comments posted and the reactions of other participants to the comments.

Implementation of the BBFG

If You Build It, Will They Come?

As described above, the BBFG discussion group guide required intense preparatory anticipation of participant responses. Every question, phrase, image, and uploaded video was scrutinized for appeal, functionality for a BBFG, and potential to expand the conversation. Table 11 provides an overview of topical themes for the BBFG, with a few examples of the questions posed for each topic.

Table 11

Bulletin Board Focus Group Topical Themes and Question Exemplars

Topics	Question Exemplars
Getting to know you	Lets watch this video called "Talk About Change" made by some young adults in Britain with life limiting conditions. The young adults in this video address lots of the topics we will be discussing. How are your experiences similar or different to what you have viewed? Tell us why.
Managing your medical condition	Thinking about your condition, In the next 2 years do you expect it to stay the same, improve, or decline? Please describe the expected changes.

Paving the Way You are all pioneers and leaders because you are among the first to live with your condition into adulthood.

If you were asked to come to a Teen Camp to help teens think about getting older with your condition, what would you tell them the most important things to keep in mind would be?

Hopes for the future Being among the first with your condition to live into adulthood, what do you hope will be different for kids who are 10 years younger than you?

Transition Changes Lets say that you are in a position to design a brand new health care program for people with a similar condition to yours.
How would it work?
Explain why you chose the aspects of the healthcare program that you did.
Take a look at other people's program ideas and tell us what you think.

Getting what you need We have been talking about the change in resources and support when you move from pediatric to adult care and the negotiating and advocacy involved to get what you need.

Who is doing most of the negotiating or "fighting" for what you need? You? Parent? Someone else?

Do you have anyone you can count on when things get really hard?

Relationships

On the video we saw the first day, one person said, "when they ask questions about me, they don't ask me, they ask the person I am with". A young woman with a complex condition told me that her wheelchair and equipment gets in the way of people seeing her as a person and knowing her for who she really is. Amy mentioned the other day that people don't expect you to be smart enough to go to university.

How are your experiences similar or different?

What do you think people would find the most surprising or interesting about you if they could see past your condition or being in a wheel chair?

Post a picture or song, or whatever would represent what you wish people would really know about you instead of your wheelchair and/or condition.

Decision
Making

What do you consider the toughest decision that you have ever had to make?

Tell us about what made this the toughest decision?

Who (or what) supported you in the process of making this decision? Or was it something you decided on your own?

Computers and “GAMING IS MY LIFE”.

Technology How true is this about you?

Feel free to elaborate on your answer in the textbox below.

Advanced Some of you will have already thought about advance

Directives directives (making sure other people know what you want done as your health declines) and some of you may not have.

Answer these questions as best as you can.

Do you have advance directives in place?

If you do please tell us what your wishes are for your advanced directives

After vetting the BBFG and completing recruitment, the audit trail review revealed my anxious wait for the BBFG to go live. With all components of the project virtual and online, pre launch uncertainty was high, wondering if participants would “show up” online, how the group would establish rapport, and if they would remain engaged for five days. The complexity of the participants’

disabilities also created uncertainty about how much encouraging and “soft sell” was appropriate to ensure their engagement.

BBFG Live!

As expected, some participants were more conversational and provided more responses and contextual information than others. The two BBFGs produced an average of 220 participant posts over 5 days, yielding approximately 11 posts per participant per day.

Table 12

Bulletin Board Focus Group Participant Response Rates

User	Total # of Posts	Average # of Completed Questions	Available Questions	Average % Completion of Available Questions
BBFG 1	193	36	38	95
BBFG 2	244	37	38	97

I encouraged the less conversational participants to add more to the discussion through follow-up probing questions and an email alert that reminded them to go back to the discussion to answer further questions. The most conversational participants tended to be the most responsive to further probes. Knowing the possibility of learning disabilities in some participants, I took care in determining the number of probing questions that should be asked, and whether to be chatty and conversational or be succinct, limiting the amount of content.

For those who had limited capacity to provide fulsome answers through text, the webcam provided another venue to participate. However, some of these participants told me that they did not think they could speak clearly enough to be understood. Others did not have an operational webcam, or else participated in the discussion using mobile device not equipped with webcam capabilities.

I also found the time commitment a factor in keeping the conversation animated, both for the participants and myself. No participants withdrew or stopped coming to the discussion, but on day 3 of 5, there was a noticeable dip in the amount of conversation and responses to questions. In addition to this mid-week lull in sticking with the daily commitment to the group, later discussion topics concerned relationships and intimacy, so participants might have experienced more difficulty forming responses. As moderator, the time commitment was very flexible; it proved easy to check in on the conversation and respond from a computer or mobile device. However, as in email correspondence, much more time was required to craft responses and phrase questions in text rather than in conversation. Especially with the sensitivity of topics about end of life, intimacy, and planning for the future with pedLTC, it was imperative to ensure that words on the screen were neither stark nor misinterpreted.

I employed several strategies to maintain animated participation. First, I rewarded desired behaviour, for example thanking participants publicly when they commented on each other's posts or gave detailed answers. Second, using the webcam made it possible to speak conversationally and personally with the group,

initiating further conversation. Third, I thanked participants twice daily for their participation on the welcome and wrap up screens, emailed an alert about follow-up questions, and when possible, referred to participant comments from previous days. Finally, as described above, I utilized a variety of question types, pictorial representations, vignettes and videos to keep participants engaged in the discussion (Van Patten, 2011).

The young adults with pedLTC who participated in this BBFG were positive about using this forum for research. For example: “I liked the variety of the topics and the way the responses were text or video. I liked the way the website is set up with profiles, and how your notified of new response so it's easy to answer.” Another said, “Some of the topics really made me think and made me question some topics that I didn’t think about before.” The BBFG was more than a method of collecting data. It created a networked community that provided connectivity with peers to learn from each other, and share resources and common understandings. After the formal BBFG discussion had ended, the participants continued to check back for more conversation. Their recommendations for change or improvement included, “I think it would be cool if these discussions lasted a little longer. And also if they’d been a larger group, so that we could really get discussions going.” Participants stated they would be very interested in participating again, and that they would highly recommend this BBFG to someone else.

Future Considerations

This BBFG demonstrates a new and novel research method to give voice to a medically fragile population with limited and divergent communication abilities. One of the greatest benefits of a BBFG is that data collection becomes an intervention. Questions and discussion amongst the group facilitates new ways of thinking about issues important to the participants. Travel time and costs for the researcher to visit participants individually, or for participants to travel to meet for a face-to-face focus group would have been prohibitive to conduct in any other format except a mail out questionnaire. In the face-to-face interviews conducted to develop the BBFG, interviews were often delayed because of fatigue and changes in condition. In addition, the face-to-face participants fatigued easily when answering a series of questions.

The limitations of this BBFG were similar to those identified in the literature: uneven participation by group members (Kralik et al., 2006), diminished group interaction dynamics (Kenny, 2005; Clapper & Massey, 1996), difficulty or lack of interest in following conversational “threads” (Moloney et al., 2003; Stewart & Williams, 2005), short responses because of limitations with typing and/or problems using webcam, and limited means for gathering contextual data (Kralik et al, 2006; Lovejoy, 2009) from those using text responses exclusively. Unlike a face-to-face discussion, when a BBFG participant does not respond to further questioning, it is not clear whether the participant ignored and/or did not know that further questions were posted, or if they had

nothing more to say. Developing the online discussion and maintaining presence and fostering discussion on the discussion were time intensive.

Some challenges identified in the literature did not arise in this BBFG. For example, researchers have been cautioned that the use of non-standardized computer and internet jargon can be confusing (Im & Chee, 2006). I found no such instances in this BBFG; likely, the universal acceptance and understanding of abbreviations common in text messaging has made them less confusing. Participants used CAPITAL LETTERS, varied font colors, uploaded pictures, and emoticons to express both significant and subtle points. The proliferation of text communication through text messaging and social media sites such as FacebookTM has “normalized” text communication and appears to have nearly usurped face-to-face communication in adolescents and young adults. With the rapid advance of technology to provide sophisticated web-based communication for business and academia, earlier logistical issues that made it difficult for participants to log in, find the discussion or maintain connectivity throughout the BBFG (Im & Chee; Moloney et al., 2003) were no longer relevant. Finally, it proved unnecessary to eliminate any potential participants because of a lack of access to computers.

Table 13 summarizes the merits and detractors of the BBFG process for researchers and medically fragile populations.

Table 13

Comparison of Advantages and Disadvantages of Bulletin Board Focus Group Process for Researchers and Medically Fragile Populations

Research process	Advantages	Disadvantages
Participant selection		
Researcher	Increases accessibility and pool of participants Increases response rates ^a	Slow responses to email recruitment due to physical limitations or not being email users
Participant	Easy to decline through no response or email Reduces isolation and creates community ^b	Inaccurate email address results in missing opportunity
Data Collection		
Researcher	Reduced time and costs Higher retention rates Enhanced theoretical saturation Control over methodological rigor of data collection Enhanced participation and easier discussion flow about	Unclear if theoretical saturation achieved when no response to further questions

sensitive topics^c

Participants	Flexible timing to respond within their time zone, and work, family and leisure commitments	Feel alone when unsure how to answer a question
	Data collection is an intervention facilitating new ways of thinking	Time commitment required to log on every day
	More time to reflect and answer at their own pace	
	Text format facilitates a concise discussion	
	More interest in participating because the format is novel ^d	

Data Analysis

Researcher	Automatic and accurate verbatim transcripts	Interface difficulties between BBFG
	Text and video analysis supported within BBFG	transcript and NVivo software
	Transcription costs, time and	

errors are eliminated

More potential for theoretical
saturation^e

^a Ahern, 2005; East et al., 2008; Fox et al., 2007; Im & Chee, 2006

^b Beck, 2005; East et al., 2008; Fox et al., 2007

^c Cantrell & Lupinacci, 2007; Courtney & Craven, 2005; East et al., 2008; Fox et al., 2007; Im & Chee, 2006

^d Abbott, 2011; Ahern, 2005; Courtney & Craven, 2005; Fox et al., 2007; Moloney et al., 2003

^e Ahern, 2005; East et al., 2008; Fox et al., 2007

Conclusion

This paper describes one of the first attempts to employ an innovative research method to access an otherwise inaccessible population. The proliferation of social media, online discussions and text conversations makes this medium familiar and comfortable for young adults, and the diversity of physical limitations and communication modalities among the participants in this study demonstrated the inclusiveness and adaptability of an asynchronous BBFG forum. In addition to the technological capacity of the online format, the quality of data generated remains dependent on the skills of the researcher and moderator in building an engaging, personalized and coherent discussion over several days using varied mediums. Continued refinement of these methods with more groups of hard to reach participants, more feedback about participants' experiences, and further understanding of the essence of the online discussion that can extend its purposes beyond a method to an intervention, will continue to add merit to BBFG methodology.

Notes

The terminology to describe young adults with pediatric life threatening conditions (pedLTC) varies among authors. Some choose to use pediatric life limiting conditions (pedLLC) and others choose pediatric life threatening diseases (pedLTD). I chose to use pedLTC to distinguish this population from 1) pedLLC which has been used to describe youth with conditions that may be life limiting, but with expectations to live well into their 50's and 60's, and from 2) pedLTD which does not reflect that many of these young adults do not have a specific disease, but rather complex conditions with multi-system affect.

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INTRODUCTION TO CHAPTERS 4 AND 5

Chapters 4 and 5 are comprised of papers prepared for publication. At the time of writing, Chapter 4 “Up Against the System” has been accepted, and is in press with *Nursing Research and Practice*, and follows the formatting required by this journal. At the time of writing, Chapter 5 has not yet been submitted to a journal. Both chapters 4 and 5 focus on the following study questions:

1. What are the experiences (opportunities and challenges) of young adults with complex/pedLTC who have transitioned from pediatric to adult services?
2. What are the most significant supports and factors that will contribute to achieving their developmental goals?

Chapter 4, “Up Against the System”, describes the barriers and facilitators in the health, education and social systems that the young adults with pedLTC experienced during their transition years. Despite the shortcoming of system and environmental supports, and the burden and impact of their conditions, these young adults demonstrate uncharacteristic resilience and hopefulness. Chapter 5, “Investing in the Future” explores the concepts of resiliency, vulnerability, and future thinking; describes how young adults with pedLTC invest in their short futures; and provides example of environmental supports and barriers in their quest to dream and achieve their goals. The study propositions overlap and guide both papers. The propositions are provided here for review:

1. Development of comparable adult medical and support services did not follow interventions to increase life expectancy (Brown, Katherine, Allen, Quach, Chiu & Bialystok, 2010; Hamdani, Jetha & Norman, 2011).
2. Health care transitions are embedded with the broader health care system and connected to the family, social, educational, cultural, religious and many other systems (Brown et al., 2010; Hamdani et al., 2011).
3. Youth with pedLTC are potentially juxtaposed between the hope of achieving new young adult aspirations and their own end of life trajectory (Doug, Williams, Paul, Kelly, Petchey & Carter, 2011; Freyer, 2004; Melies, Sawyer, Im, Hilfinger & Schumacher, 2009).
4. A continuation of the integrated palliative care services (including medical, psychosocial, educational, spiritual and social needs) received from 0 – 18 years is not available to young adults with pedLTC in B.C. and this could result in increased morbidity, isolation, and inadequate support to fulfill their aspirations for intimacy, independence, education, work and interpersonal relationships (Meleis, Sawyer, Im, Hilfinger & Schumacher, 2000; Prestige, Romann, Djurdjev & Matsuda-Abedini, 2012).
5. Health and social service providers in the adult care sector could have limited awareness of the complexity of the issues for youth with

pedLTC, or do not have a mandate, or the resources to provide integrated services to young adults with pedLTC in B.C.

6. Current transitional services from integrated pediatric services to the adult health and social service sectors are limited in their breadth, scope and effectiveness to meet the needs of young adults with pedLTC (McDonagh, 2006; American Academy of Pediatrics, 2011).
7. Vulnerability; personal factors, such as cognitive capacity, temperament, self direction, imagination, values, and meaning of transitional phase; parental influences, such as availability, history of acceptance to youth decision making, attitudes or preoccupation with youth's strengths or limits, view of impact of youth's pedLTC on siblings and family; and external supports such as extended family, teachers, community, religious, and cultural mentors can influence the youth's experience of the transition process (Suris, Michaud & Viner, 2004; Ungar, Brown, Liebenberg, Cheung & Levine, 2008).

CHAPTER 4

UP AGAINST THE SYSTEM:

A CASE STUDY OF YOUNG ADULT PERSPECTIVES TRANSITIONING
FROM PEDIATRIC PALLIATIVE CARE

INTRODUCTION

A life threatening condition adds complexity to the challenging period between childhood and adulthood and the needs and experiences of young adults are specific and different from children's and adults needs [1]. Unfortunately, advances in pediatric care have not provided the interdisciplinary support services required by those young adults with pediatric life threatening conditions (pedLTC) who live beyond childhood, but have limited expectations to live past early adulthood [2, 3, 4]. These young adults, who are the first generation with pedLTC to live into adulthood, face a multitude of challenges transitioning from the plethora of pediatric services to scant adult health services [5] and are vulnerable to a significant deterioration in health status and even increased mortality once they leave pediatric care [3, 6]. While the number of young adults with pedLTC is small, the numbers continue to rise [7]. In the United Kingdom, the number of 16-19 year olds living with life threatening conditions nearly doubled in a decade [1]. To date, there is a paucity of research about the transition experiences of youth "aging out" of pediatric palliative care [3].

Palliative care is an interdisciplinary approach to the management of incurable or life threatening diseases [8]. However, the philosophical differences between pediatric and adult palliative care exclude this population from adult palliative care services because pediatric palliative care is lifelong until death, whereas adult palliative care focuses on the last weeks and months of life [7, 9]. The differences in definition and types of services offered in palliative services for youth and adults have resulted in confusion and abrupt ending of supportive services for young adults with pedLTC [10]. Currently, young adults with pedLTC live in a zone of disability and chronic illness that has no specific care delivery system [11].

Background: The Transition Experience

While not specific to young adults with pedLTC, the transition literature for youth with complex conditions does provide insights about this complex phenomenon. Transition is the planned and purposeful movement of youth with chronic medical conditions from comprehensive pediatric services to adult-oriented health care [12]. Preparatory transition work occurs between 12 -18 years of age, but the real lived experience of the transition occurs in the early adult years [13, 14]. Many comparisons have been discussed in the chronic health transition literature between pediatric and adult healthcare systems, which also apply to the experiences of young adults with pedLTC. In the pediatric system, the health care team and parents make decisions with, or in the best interest of young people. In adult care, young adults are expected to navigate new systems and make their own decisions [15, 16, 17]. This approach does not consider the impact of the young adult's cognitive and verbal abilities, patterns of family decision-making, adjustments to their condition, and other factors that may leave the young adult vulnerable in the adult system.

The experience of navigating the transition process is influenced by multiple and diverse factors [3, 18] such as the skills and knowledge of the youth and their families [19], the training, expertise and coordination of health care providers and services in both the pediatric and adult sectors [20], family support [21, 22] and the availability of education and social service system supports [3, 23]. To date, research in transition has focused on specific health conditions, such as diabetes, cystic fibrosis and congenital heart defects [2, 13] and on specific skill acquisition for youth and pediatric, rather than adult health care providers [3]. Further, the approach to care for young adults becomes medical-specialty focused (respirology, cardiology, etc.), rather than attentive to whole person care, which is essential for symptom management, care planning, and quality of life decision-making.

However, social and educational supports are equally important in the transition phase. A community-based approach integrating health and social services that include developmental components of young adulthood such as independent living, social, educational and career needs is recommended [24, 25, 26]. In addition to improved services in adult care to accommodate their changing health status, and learning to navigate new systems, support for the enormous psychological issues, such as the impact of their condition on themselves and their family, is also recommended [5].

For young adults with pedLTC, opportunities to practice taking on the responsibility of their own health care can be limited because of repeated and prolonged hospital stays, intrusive home care regimens, dependence on others for care, learning disabilities, sporadic school attendance, and fewer socialization opportunities [5, 27, 28]. In addition, for young adults who are cognitively capable, the continuation of family centered care through adolescence limits their opportunities to develop autonomous health self-management skills [11]. For young adults with pedLTC, learning to navigate adult systems early in the transition process is important.

Finally, pedLTC are rare and unfamiliar in adult care. This has resulted in a dearth of experienced adult clinicians, and interdisciplinary care groups that understand and have the resources to manage this group's complex care needs [12, 29]. Adult general and specialist health care practitioners may be unwilling to accept a youth with pedLTC into their care because of the time required to manage the complexities of the youth's condition, as well as their own unfamiliarity and lack of expertise with these rare pediatric conditions [13, 30, 31, 32]. Primary care providers might have only one pedLTC patient in their entire career and adult specialists are not trained in pediatric conditions [30, 33]. Therefore, for young

adults with pedLTC there is no real system to accept them after pediatric care [11].

METHODS

Research Design

This paper is part of a larger descriptive case study, guided by Yin [34], that explored 1) the experiences of young adults with pedLTC who have transitioned from pediatric palliative services to adult care, the significant supports and factors that contributed to, or were barriers to achieving their developmental goals, and 2) the advantages and disadvantages of an online focus group to engage medically fragile participants. Propositions guide case study data collection and provide boundaries for analysis. The propositions for “Up Against the System” are listed in Table 14.

Table 14. Propositions: Up Against the System

1. Youth with pedLTC are potentially juxtaposed between the hope of achieving new young adult milestones and their own end of life trajectory.
2. Health care transitions are embedded with the broader systems and are connected to family, social, educational, cultural, and religious factors [3, 23].
3. Development of comparable medical and support services did not follow interventions to increase life expectancy [3, 4, 5].
4. Current transitional services from integrated pediatric services to the adult health and social service sectors are limited in their breadth, scope and effectiveness to meet the needs of young adults with pedLTC [16, 20]. This could result in increased morbidity, isolation, and inadequate support to fulfill their aspirations for intimacy, independence, education, work and interpersonal relationships [6, 21].
5. Resiliency, vulnerability, personal factors (cognitive capacity, temperament, self direction, imagination, values, meaning of transitional phase, and motives), parental influences (availability, history of acceptance to youth decision making, attitudes or preoccupation with youth’s strengths or limits, view of impact of youth’s pedLTC on siblings and family), and external supports (parents, siblings, extended family, teachers, community/religious/cultural mentors and volunteers) can influence the youth’s experience of the transition process[21].
6. Health and other service providers in the adult care sector could have limited awareness of the complexity of the issues for youth with pedLTC, and/or do not have a mandate and/or the resources to provide integrated services to young adults with pedLTC [30, 31, 33].

The objective of this paper is to describe the complex interplay of the health, education and social service sectors in the transition experiences of these young adults. Accessing this population with traditional qualitative interview methods,

such as focus groups, is difficult because their numbers are small; they are geographically dispersed; the complexity of their conditions requires many supports such as care attendants for personal care and respiratory support, equipment such as power wheel chairs and computer assisted communication devices; and they have divergent mobility and communication abilities related to the impact of their conditions. An innovative method of data collection to overcome these barriers is described below. Two university ethical review boards approved this study.

This case study was conducted in two phases. The first phase consisted of three in depth face-to-face interviews with two participants. The goals of these interviews were to understand their transition experiences with health, social and educational services, and to compare their experiences with the research literature and practice recommendations on transition for youth with chronic health conditions. The second phase of this case study included the development, testing and implementation of a bulletin board focus group (BBFG), which is an asynchronous modification of an online focus group. Content for the BBFG discussion was developed utilizing the intensive one to one interviews described above, in addition to consultations with a pediatric hospice transition team, a young adult with a pedLTC who exceeded the age criteria for this study, and pedLTC experts in health and social services.

The Sample

A purposeful sample of youth with pedLTC, aged between 19 and 29 years, with limited expectations to live beyond their first decade of adulthood (29 years), having pedLTC such as Duchennes Muscular Dystrophy, Spinal Muscular Atrophy, Friedrichs Ataxia, and undiagnosed conditions, were selected from the graduates of a children's hospice in Western Canada. Within the hospice, the numbers of youth with pedLTC transitioning from pediatric hospice are very

small (8 -10 per year) and about half of these graduates met the following selection criteria: English speaking, cognitively able to participate in their care and decision-making, owning a computer, and having either verbal or typing ability sufficient to participate in face to face interviews or an online focus group. The hospice has transitioned approximately 97 youths from its program in the past 15 years. Of these, approximately half were eligible for this study. Most of these young adults require 24-hour attendants for personal and respiratory and feeding support. Their independent function may be limited to minimal movement of their index finger or a dot on their forehead to control their power wheel chairs, computers and phones. All the participants have lived with their conditions for more than 10 years, and most were diagnosed when they were less than 5 years old.

For ease of reading and protecting confidentiality, the young adult participants are referred to as “he” and all other participants as “she.”

Data Collection

Data for this two-phase study was collected over 12 months. In the first stage of this case study, two young adults with diverse and unique experiences that differed regarding characteristics such as diagnosis, age, gender, family support, opportunities for post secondary education, and living arrangements, were invited to participate in three face-to-face interviews. These participants were similar in their physical limitations requiring 24-hour care and possessing minimal movement adequate to control their power wheel chairs, computers and phones. One participant was able to speak directly with me, and the other used a computer-assisted device to type out his answers.

In the second phase of this study, a BBFG was conducted with two groups of four young adults with pedLTC. Goals were to further explore the key issues and

experiences described by the face-to-face participants, extend understanding of their range of experiences and successes and barriers within the health, education and social sectors, and provide recommendations for change from the perspectives of young adults. These participants were selected to achieve variety among their diagnoses, gender, age, and post transition experiences. The BBFG provided an opportunity to bring together small groups of young adults with pedLTC, who otherwise could not be convened. This is the first circumstance of using a BBFG to gather data from this population.

Each BBFG was conducted for five days. Daily discussions were set up by the researcher/moderator using a combination of text and video based questions. Participants logged in and out of the discussion at their convenience answering questions posted on the discussion board, follow-up questions posted by the moderator, and those posted to each other in response to other participants' comments. Participants chose text or video options to respond to the discussion questions. The discussion became interactive as the participants responded to the moderator's questions and each other's responses. The greatest strengths of the BBFG were: 1) the appeal of this methodology for young adults, 2) creating a forum discussion for young adults with pedLTC that would not have been possible due to the logistical difficulties associated with their conditions, variant communication modes and barriers and financial costs, and 3) in addition to its value as a research method, the multi day focus group became a community for the participants. Participation rate was very high with respondents answering an average of 96% of the questions and contributing to the discussion every day.

Data analysis

Face-to-face interviews with participants and experts in pedLTC were taped, transcribed and imported into the NVivo 9™ software analysis program. Transcripts from the focus groups that included all of the contextual content of the

conversations such as capitalization, multiple exclamation marks, emoticons and photos were also imported directly from the iTracks™ discussion board into NVivo 9™. Conventional and directive content analyses [35, 36] were used along with a constant comparative analytical process to review emerging patterns, themes, variations and relationships among the data sets [37, 38]. Case study analysis was directed and confined by the theoretical propositions that frame the study [34].

Credibility was ensured through use of complementary case study data collection strategies, such as the face-to-face interviews, online focus groups, and content analysis of the literature, program recommendations and professional and jurisdictional policies. These varied strategies triangulated the data sources; prolonged engagement with participants, documents and data; and facilitated persistent observations, member checking, and searching for disconfirming evidence [39, 40, 41, 42]. Specifically, the BBFG strengthened credibility through 1) high response and retention rates, indicating that participants were interested and engaged over a long period of time [40, 41], 2) the immediate generation of transcripts directly from the participants' responses, including every word and textual descriptor [43], and 3) a constant member checking, as participants post responses to both the researcher and other participants. Finally, a pediatric palliative program transition expert and a physician, and two participants from the BBFG's, reviewed the results for credibility.

RESULTS

Characteristics of the Participants

Fourteen participants were recruited to participate in this study. Two participated in face-to-face interviews. Of the remaining 12 participants, 8 agreed to participate in the BBFG. Refer to Table 15.

Table 15. Participant Characteristics for Face-to-Face Interviews and Bulletin Board Discussion Groups

n=10

Conditions	# of Participants	Age Range and Mean	Gender
Duchennes Muscular Dystrophy	4	20-28 23	Males = 4
Spinal Muscular Atrophy	3	21-26 23	Females = 3
Other: Undiagnosed, Brain Tumor, Friedrichs Ataxia	3	20-22 21	Males = 2 Females = 1

Up Against the Systems

The Canadian Pediatric Society [44] stresses that transition goals should include the adolescent’s realization of personal potential for activity, education, recreation and vocation, completion of adolescent developmental tasks and the attainment of self-esteem and confidence. Ideally, young adults with special needs can achieve a self-directed life. However, when faced with the inevitable gaps in the systems and lack of coordinated services among the health, educational and social sectors, the participants employed various thoughtful and novel strategies to get what they needed to move forward with their lives. When they came to an impasse in the education and social support systems, they found more opportunities to navigate around, or negotiate for what they needed than when they encountered barriers in the health care system. Their successful strategies included: thorough research to determine all the available services and how to access them; using their “village” for ideas (talk to others who have succeeded ahead of you) and advocacy (asking influential people, such as civic and health care leaders, to write a support letter); being persistent, positive and creative; and never taking no for an answer. In some circumstances, the systems were too formidable to impact, and they were stalled or unable to move forward.

The following section describes what the young adults in this study most want from the health, education and social systems: knowledgeable providers, coordinated and accessible services, being respected and valued, and services and supports that promote their independence. System deficits and strengths, and the young adults' resilience and coping strategies to overcome the deficits to get what they need to move forward with their lives are also described.

Knowledgeable providers. Participants had varying levels of satisfaction with their health care providers. The participants referred mostly to their physicians because they did not have access to multidisciplinary health team members in the adult sector. For this population, access to individual adult specialists who had comprehensive understandings of their condition was difficult; access to a team of physician experts was impossible. Many found that their physician's knowledge about their pedLTC was limited, and they were educating their doctor about their condition. As one of the young adults explains, "*it's been really challenging for me because not only do I have complex health issues, but I have to educate my doctors about what (my condition) is.*" Further, even adult specialist palliative care was not a supportive experience because of lack of expertise in pedLTC and young adult developmental goals.

Without understanding of these pedLTC, health care providers were unable to provide anticipatory guidance and management strategies. Reactive rather than proactive care was one of the most significant changes and struggles described by these young adults. One participant commented,

When I was a kid it was like the doctors kept on top of my medical condition and everything involving it. In adulthood I really have to seek out any sort of help when it comes to specialists and doctors.

Compared to their solo adult practitioners, their multidisciplinary pediatric teams were perceived as "very active" with information beyond health care needs that

included housing options, equipment, and support for education. When condition specific expert care was not available, the young adults remained in acute care settings for extended periods of time, or received standardized medication protocols for the “normal” population that resulted in untoward side effects and consequences for young adults with pedLTC.

Knowledgeable care was also compromised by ineffective communication among care providers. This added stress for the youth, or in some cases, family members or their pediatric health team, who had to advocate, update and educate each of their new adult health care providers. Some young adults were unable to take on this responsibility and bridge the communication gap among their care providers because of the complexity of their condition, resulting in declining communication abilities, and access barriers such as re-referrals.

When their conditions were stable, participants were satisfied with the knowledge of their health team and access to the care and services they received. They were empowered by the experience of taking control and being their own advocates. These participants had not experienced any recent changes in their conditions and while they were stable, they did not “*need a team hovering over me all the time.*” However, they were aware that for their friends whose health was not stable, access to care was difficult and not easily coordinated. While all participants acknowledged that their health status would decline, they were overwhelmingly hopeful and positive that their conditions would remain stable for the next 5 - 10 years. Anticipating a decline in their condition in the future, they indicated that they would prefer an expert team of health care providers, similar to their pediatric palliative care experiences, including specialist nurses. Only a few participants had experience watching a friend die with their same condition, and most participants had not planned for their declining health, preferring to focus on their current stable condition and hopeful that it would remain stable, or that new

technologies and cures would be discovered before they faced declining function and symptoms.

Coordinated and accessible services. During their pediatric health care, efforts had been made to coordinate appointments with their multiple care providers. This was facilitated by the proximity of physicians and multi-disciplinary team members within the same hospital. For young adults with neuromuscular type pedLTC in the adult system, the organization of separate appointments with their primary care physician, specialists in respirology, neurology, cardiology, gastroenterology, physical and occupational therapy, and nutrition, medical and oxygen suppliers becomes daunting. Getting to doctors' and service providers' offices is a Herculean effort, requiring special transportation accommodating their power wheel chairs, organization of their supplies and equipment, and availability of parents, partners, or care aides to attend the appointments with them. Required repeat visits to their family physician to access their specialists added even more appointments and logistical problems, as described by this participant:

It can be really annoying because like every six months if you don't go see your specialist then they close your file and you have to get a new referral each time. And for a complex . . . disease that will go on for my whole life, like it's not a temporary thing. . .

Accessibility to health services was improved when there was prompt coordination of care among their physicians. Participants who were satisfied with their access to health services described a coordinated effort between their primary care provider and adult specialists, which resulted in quick and efficient response to symptoms and subsequent treatment in hospital.

Reliable and prompt access to health care services can be problematic, especially for those young adults living independently or in extended care facilities. For

example, when a participant's call bell was not answered and he required suctioning, he "face booked" his friends and asked them to call the nurse's desk at the extended care facility to tell them that he needed help right away. In another example, a participant was unable to travel by ambulance to hospital because lying down was too painful. He was transported to the hospital emergency room in a police "drunk tank van" that accommodated his power wheelchair and returned to his facility via an out of service city bus. While these examples are unusual, it does demonstrate the significant gaps in services and safety that arise for young adults with pedLTC who do not have a dependable network of support.

Problems with access to academic and social support programs were universal. Participants emphasized the need for an integrated program that provides seamless resources and supports between these two systems. For example, for those attending post secondary education prior to turning 19, many experienced a one-year gap between the end of funding from the pediatric program and the start of adult funding, resulting in inadequate resources for both home and school support.

In addition to coordination gaps between the systems, accessibility to post secondary education was hindered by their high schools' and post secondary counselors' lack of knowledge about the grants, programs and care aid funding available to support their education. For the participants attending post secondary institutions, accessing the resources they needed required lobbying the student assistance departments that were intended to support them with funding and accommodations. Asking influential pediatric health care providers to write a letter on their behalf was one successful strategy that secured necessary support. For example, one participant who fought to have bathroom accommodations in place so that he could attend university said: *"I think that my advisor decided to change her mind because there were some people advocating with me and she*

saw that there was a need, and if she didn't install an accessible washroom for me it would be considered discrimination.” If lobbying efforts were not successful, or if the participants did not have personal resources to pay for tuition, accommodative renovations to post secondary facilities, and attendant's wages, education plans were not achievable or were postponed. Even while their conditions were stable, fatigue, sickness and illness and communication restrictions impacted their ability to achieve their post secondary goals. Further delays to secure the necessary supports could mean shortening, or preventing the pursuit of their educational dreams as their health condition further changes and deteriorates.

Only one participant who attended a post secondary institution described receiving sufficient support, resources and guidance.

Being respected and valued. In addition to improving the delivery of services in the health, education and social systems, being respected by the providers of these services was also highly valued. For example, health services focused on disability rather than ability were viewed as condescending:

The doctors who specialize in 'normal' things sometimes look at me like I have a disability and I deserve to be treated differently because of that.

They've not been very understanding that we all just want to feel and live as normal a life as everyone else.

While young adults with pedLTC are disabled, normalization in their lives is important, including interactions with their health providers.

The participants attending post secondary education experienced being devalued as potential students and being questioned about their cognitive capabilities because of their disabilities. One participant was told that his “*needs were too intensive*” and many of the students related that when they asked their professors

questions about their class, the professor spoke to their care attendant instead of them. Patronizing and condescending attitudes that felt demeaning and inconsistent, and incorrect information about resources and services for which they were eligible, limited their access to and funding for post secondary education.

Opportunities for independence. As the first generation of young adults with pedLTC to live into adulthood, these young adults are pushing the current system boundaries to get the supports and resources they need for independent living. Their paradox is that gaining independence necessitates being dependent. Their community of caregivers, family and friends, and their equipment and supplies are their means to independence. As one of the participant describes, “*there are some things I need assistance with, and even though I'm super stubborn, these are things I just have to accept.*” For those with a tracheostomy, an attendant is necessary 24 hours a day. However, for some, short periods of being alone are worth the risk of the tracheostomy becoming dislodged and suffocating without help. Recognition and acceptance of their dependence on physical support in no way diminished their strong resolve for independence.

Of all the system-focused struggles identified by these young adults, achieving sufficient support for independent living was the most difficult. Acquiring sufficient attendant time and resources from the program designed to support their independent living required sophisticated research and advocacy skills. While all of the participants hoped to have an opportunity to live independently, efforts to procure funding support for care attendants and equipment such as specialized beds and computer assisted technology for safety, plus the cost of renting accommodations suitable for their power wheel chairs and other equipment, was not only frustrating but in most in most cases insurmountable. One of the 10 participants was living independently during data collection, partly made possible

by having a spouse. Another participant was in the process of lobbying for support to move from an extended care facility to his own apartment. Thirty-five care providers and managers from the health and social service sectors attended a meeting to discuss resource allocation and coordination for one person's move. Equally frustrating for participants were either experiencing "no expert availability" or dealing with too many experts who did not coordinate their roles and services.

Some young adults with pedLTC live in extended care facilities because they do not have the support to live at home or independently. Here, it was found that young adult developmental goals and desire for independence clashed with the culture of extended care. Even for those who lived by a mantra of "*never give up fighting*," opportunities to achieve their personal goals were constrained and deflated by maintenance style care that did not encourage or support their independence, or education, recreational and vocational goals. For those who are not "fighters," hopefulness became hopelessness.

Making the Systems Better

Young adults with pedLTC, having grown up in the pediatric health care system and experienced the challenges of transition and adapting to an adult health care system that was not designed nor equipped to meet their needs, can offer invaluable perspective to practice and policy leaders regarding how to improve the systems.

The young adults in this study recommended a team approach to treating their medical condition, an extended transition period over five years that facilitated consultation between their pediatric and new adult providers, access to a multidisciplinary clinic of experts related to their conditions, one time referrals to specialists that would not require repeat visits to a family physician, and more

communication between their family physician and adult specialists to keep their family physician apprised of their care needs. They also hoped for medical and technological advances that would continue to improve their health status.

Desired improvements to the education system included more funding support for tuition and their personal care attendants. A forum in which people transitioning with disabilities could learn about the resources available at each post secondary institution, how to access these services and how to lobby for supports not currently available were also recommended.

In the social services sector, their suggestions included changing the age at which adult funding started to coordinate with the end of pediatric funding, and increasing eligible hours of care to support independent living. One participant recommended creation of a service that he would call “HandicApparel,” from which medical equipment could be accessed for reasonable prices. Access to a place to get away (similar to pediatric hospice) with support for their personal care, end of life care, and opportunities to socialize with other young adults with similar conditions were also proposed.

DISCUSSION

While there has been extensive research, description and opinion reported on transition challenges and solutions for youth with chronic health conditions, this study is among the first to describe the unique transition experiences for young adults with pedLTC. The participant data exemplifies the multiple intersecting factors that influence the transition experiences of young adults with pedLTC: their individual characteristics, such as skills and knowledge for self or directed care; interpersonal support from their family, friends and community; organizational coordination and support between pediatric and adult services; and

policy resources to support transition [3]. Further, the participant responses demonstrate the creative and time consuming work the young adults, their families, volunteers and professionals have undertaken to overcome system barriers and initiate changes that need to be normalized across all the health, education and social service sectors.

System Improvements are Necessary

The current health, education and social systems do not fully support the transition of youth from pediatric to adult services. The complexity of the medical transition process has resulted in a fragmented focus on health care transitions that mostly exclude the social and educational systems [3, 23, 25]. However, even if well integrated, health care services do not create a healthy community [25]. For example, education is one of three strongest determinants of health and well being for adolescents and young adults [45], but the young adults in this study faced barriers and prejudice accessing post secondary education. Successful transitions require policy approaches that move beyond health care needs and recognize the full range of young adult developmental needs [46]. Cross system linkages are necessary to facilitate timely and efficient support between the pediatric and adult health, education and social systems to support and enhance these young adults' limited remaining years. Most importantly, modifications to current practices and development of new services and programs should be informed by the views of the young people themselves.

From a systems perspective, there is no system for young adults with pedLTC to transition to [3]. Adult health, education and social systems do not coordinate or integrate their services, and have do not have central leadership, resulting in mismatched programs with differing intentions. Health care system transition includes the interacting stakeholders (young adults, family members, health care

providers, administrators and policy makers); pediatric and adult health care providers and organizations; and the specific transition interventions [3].

Bridging the chasm between pediatric and adult health care is especially complex for young adults with pedLTC. First, prior recommendations for improving transition from pediatric care to adult care such as 1) starting preparation for transition in the early teen years, 2) specific guidelines to ensure that requisite skills and knowledge are achieved prior to transfer, 3) training of adult health care providers, and 4) ensuring efficient transfer of comprehensive health and personal information [12, 26, 47] remain unattained for young adults with pedLTC and their providers. Further, many of these young adults receive all of their health care through their pediatrician. When they reach adulthood, their adult primary care provider is not familiar with their health needs, coordination of services, and preferences. Ensuring that youth are seen by their primary care providers throughout their specialized pediatric care, and improving interconnectedness between pediatric and adult services, may create accountability for young adults dropping out of adult care, and accelerate adult services to adapt to the needs of young adult patients [61].

Second, symptom management for young adults with pedLTC becomes more complicated in adult health care because of the broad array of rare and unknown conditions, and few corresponding opportunities for adult health care providers to acquire specialized knowledge. Without access to supportive health services, young adults lose hope and power waiting months to see an adult specialist for symptom relief. Moreover, pedLTC in young adults often follow a series of declining plateaus in health status that are punctuated by periodic life threatening symptoms with little predictability about which crisis event will be life ending [48, 49, 50, 51]. With limited and unknown time remaining in their lives, waiting

months for symptom relief has a significant and detrimental impact on the quality of their life.

Third, previous research has found that adolescents with chronic illnesses were less likely to want to discuss their end of life preferences than those who were healthy [52]. Therefore, expert knowledge about symptom management and end of life trajectory, a trusting relationship with adult care providers skilled in end of life planning, and a coordinated plan for medical emergencies, are essential components of health system support for young adults with pedLTC [53].

Fourth, better access to coordinated physician services will require a concerted effort among physician stakeholders to effect changes within their billing regulations. For example, billing practices that allow patients in the transition years (18 - 30) with specific conditions to be seen as needed by the appropriate health care provider for changing symptoms without re-referral every six months will not only improve ease of access, but also decrease morbidity symptoms [6].

The complexity of navigating multisystem changes with complex and life threatening conditions requires a specific kind of health care professional and health services. Until recently, there has been little focus on primary care providers in the transition process [54]. The role of a primary care home is now prominent in clinical guidelines on transition [29, 46]. Primary care philosophy recognizes that health transition must also include education, career choices and independent living [46]. In pediatric care, nurse clinicians provide primary care management of complex youth and families in tertiary care settings. In adult services, there is no “safe passage” for young adults to an equivalent supportive care coordinator. Nursing experts understand the unique health, developmental and social challenges of this population, and are well suited to provide ongoing support for the coordination of future health care, as well as liaison for education

and social services planning [53]. A joint report from the Canadian Health Research Foundation and Canadian Nurses Association (2012) have determined that community-based and nurse led interdisciplinary teams that include a primary care physician are key components of effective and efficient provision of complex care to a complex group of patients [62]. Multi-faceted interventions that include both health and social interventions require specially trained or advanced practice nurses to supplement primary care physicians and other health care professionals. This coordinated model of care provides proactive and comprehensive community care to vulnerable populations with complex health conditions and social circumstances [62].

There is evidence of specialized resources for young adults with pedLTC in the United Kingdom where children's hospices have expanded their upper age limit into the young adult years, and new young adult specific hospices and hospice societies are being developed [1, 55]. Like pediatric hospice, young adult hospice (YAH) is philosophically different from adult hospice and can function as an episodic primary care home. The goals of YAH include anticipatory guidance about managing changing symptoms and planning for end of life through the entire trajectory of life, not just the last weeks and months. Young adult hospice homes and hospice societies can also encourage successful transition from pediatric to adult services, provide specialized symptom control, psychosocial and spiritual resources, support independence, social networks, recreation, end of life decisions and care, and access to work and educational opportunities. Hospice services for family members can include respite and bereavement support [2, 53]. Young adults living in non-urban areas can be supported through telehealth symptom management, the expert support of a network of providers, and information about how to facilitate online social connections and navigate multi-sectoral services in their area. The input and voice of young adults in the development of hospice services is imperative, to ensure hospice services are

responsive to their needs and life goals [1]. Engaging young adults with pedLTC in the development and ongoing support of hospice social networks, and providing peer support about how to access health, education, vocational and independent living resources, will create meaningful work, volunteer and social opportunities.

STRENGTHS AND LIMITATIONS

While this is a small study and the results are not generalizable, it is among the first research studies to document transition experiences from pediatric to adult services from the perspective of young adults with pedLTC. Additionally, data was collected using an innovative method, the BBFG, to reach a medically fragile population. While there were instances of limited follow-through by some participants on the BBFG discussion threads, the resulting data was rich and creative. Further research opportunities include investigation of transition experiences for young adults with pedLTC from the perspectives of pediatric and adult care providers, and family members. Developing collaborative training and sharing of resources between pediatric and adult providers and programs will improve the transition experience for young adults with pedLTC.

CONCLUSIONS

After nearly a decade of effort toward implementing programs, protocols and policies to improve transition from pediatric to adult care, very limited progress in new services has been made [3, 23, 29, 53]. Ideas and programs do not become diffused into normal practice without continual investment and appraisal of their status. For young adults with pedLTC, impasses continue to exist because of the history and culture of ingrained practice models, and deficient system policies and legislation for chronic illness transition [56]. These impasses are compounded by

the complexity of their conditions, uncertain and sudden changes in their illness trajectory, lack of understanding by service providers of their conditions and resources required for living, and educational and vocational pursuits, and lack of knowledgeable, supportive and trusting relationships with adult care providers [57].

Much of the transition literature refers to the transition phase in terms of disempowerment: “It feels like falling off a cliff” [17]. The loss of comprehensive services for young adults with pedLTC is profound and can be incredibly difficult without systems and family support. Despite these difficulties, all of the participants in this study were very optimistic about their future, exceeded the expectations provided by their health care providers, and anticipated that improving technologies and medications would continue to extend their lives. However, system barriers did limit and sometimes impede their efforts to live their young adult lives to their fullest, even while their health condition was stable. Resilience, community support, persistence and hopefulness enabled these young adults to navigate and persevere through system barriers [58, 59].

Care coordination through YAH programs is a substantive addition for young adults with pedLTC, who are entering adult health care with complex lifelong histories, multiple medical interventions, and varying system navigation experiences [60]. It is imperative to implement multidisciplinary and multi-agency system solutions focused on young adult priorities to guarantee that timely and seamless resources will continue to support these persons’ health, educational, vocational and social goals, in their quest to maximize their opportunities for young adult experiences in an abbreviated time frame.

NOTES

The terminology used to describe young adults with pediatric life threatening conditions (pedLTC) varies among authors. Some choose to use pediatric life limiting conditions (pedLLC) and others choose pediatric life threatening diseases (pedLTD). I chose to use pedLTC to distinguish this population from 1) pedLTC which has been used to describe youth with conditions that may be life limiting, but with expectations to live well into their 50's and 60's, and from 2) pedLTD which does not reflect that many of these young adults do not have a specific disease, but rather complex conditions with multi-system affect.

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CHAPTER 5

**INVESTING IN THE FUTURE:
A CASE STUDY OF TRANSITION EXPERIENCES OF YOUNG ADULTS
WITH LIFE THREATENING CONDITIONS**

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Introduction

Change in all spheres of life is the hallmark of young adulthood: biological, social, developmental and cognitive (Suris, Michaud & Viner, 2004). The impact of this change is described as being “alternatively exhilarating, disconcerting, satisfying, frustrating, and terrifying” (Simpson & Kettyle, 2008, p.1). For young adults with pediatric life threatening conditions (pedLTC), the burdens of their condition, disabilities, and uncertain life trajectory have the potential to both limit and intensify their experiences. Advances in pediatric medicine and new technologies have created a generation of young adults with pedLTC surviving into adulthood; however appropriate services to support them in these young adult years have not paralleled their increasing life expectancy (Hamdani, Jetha & Norman, 2011; Stewart, Stavness, King, Antle & Law, 2006). At the pinnacle of their developmental changes from adolescence to adulthood, they leave behind comprehensive, supportive and coordinated health, social and education systems, and fall into an abyss of uncoordinated adult services not compatible with the purpose, quality or quantity of the resources provided in the pediatric systems (Amari, Stinson, Cullen-Dean, Sappleton, & Kaufman, 2011; Doug, Williams, Paul, Kelly, Petchey & Carter, 2011; Lotstein, Ghandour, Cash, McGuire, Strickland & Newacheck, 2009). Consequently, these young adults

must often construct a “new normal” for their journey forward (Walsh, 2013).

This paper describes the monumental effort required to overcome health, education and social system deficits that impact transition, and coping strategies young adults with pedLTC use to invest in their short futures.

Transition Factors

Transition for young adults with pedLTC is a complex phenomenon influenced by many factors, such as: the skills and knowledge of the youth and their families (Kennedy, Sloman, Douglass & Sawyer, 2007); training and coordination of health, education and social service providers and services in the pediatric and adult sectors (Lotstein et al., 2009); and family support (Baumbusch, Lynam, Sather & Wolverson, 2009; Stewart et al., 2009). In addition to significant changes in the support and delivery of health, education and social services, young adults with pedLTC also experience developmental and situational transitions from high school to postsecondary education or vocation, and potentially from home to independent living (de Camargo, 2011; Hamdani et al., 2011; Meleis, 2010).

Given these complexities, it has been determined that health promoting interventions are most effective if 1) health and social services coordinate around a common set of goals including the health, well-being and social context of youth, and 2) health services recognize and improve services for specific populations over their life course, not in fragmented age-specific segments (Brown et al., 2010). These interventions have not been achieved to date, resulting

in two major shortcomings in the transition from pediatric to adult services: 1) services are not seamlessly connected between pediatric and adult services, and 2) the interconnectedness of health services with education and social systems is ignored (Brown et al.). Therefore, accessing appropriate health and palliative services (Doug et al., 2011), and achieving young adult developmental goals such as entering post-secondary education, finding meaningful vocation, and living independently, are fraught with complications and barriers (Young, Barden, Mills, Burke, Law & Boyell, 2009; Stewart, Gorter & Freeman, 2013) and require enormously more personal investment for young adults with pedLTC than for other young adults.

Vulnerability and Resilience

Vulnerability is influenced by the intersection between personal resources, such as emotional, intellectual and behavioural factors, and environmental supports, such as family, community and culture (Melies, 2010; Rogers, 1997). In addition to the complications of uncoordinated systems during transition from pediatric to adult services, transitional periods intensify vulnerability and increase the risk of poor health outcomes (Meleis, Sawyer, Im, Hilfinger & Schumacher, 2000; Prestige, Romann, Djurdjev & Matsuda-Abedini, 2012). While vulnerability is inherent in human experience (Walsh, 2012), for young adults with pedLTC there is a potentially synergistic multiplying effect of factors that can increase their vulnerability, such as: changing and declining health; profound physical disabilities; debilitating pain; dependence on care providers and

equipment for daily living; feeling isolated; the psychological burden of their life threatening condition for themselves and their family; uncertainty about dreaming or achieving goals; and navigating and negotiating new systems (Meleis et al.; Simpson & Kettle, 2008; Suris et al., 2004).

However, vulnerability is not synonymous with weakness, and can be transformative with supportive resources and protective personal factors such as cognitive abilities, easy temperament, self-regulation and a positive outlook, (Supkoff, Puig & Sroufe, 2012; Walsh, 2012). The cognitive appraisal process (Lazarus & Folkman, 1984) describes that under comparable situations, the interplay of environmental and personal factors results in varying responses among people, and at different times for one individual. Some will feel angry; others depressed, anxious or guilty; and others will feel challenged rather than threatened. Further, while the impact of their condition pervades the life experiences of young adults with pedLTC, it does not take center stage while their health is stable (Stewart et al., 2013; Fletcher Johnston, 2012).

Resilience is conceptualized as an unfolding developmental process that harnesses psychosocial, environmental, biological and cultural resources to sustain well being (Panter-Brick & Leckman, 2013). Studies on youth resilience have investigated the impact of environmental factors, such as parental mental health and behaviour, divorce, abuse, social disadvantage, and war (Rutter, 2013). Resilience is a valuable lens through which to explore the impact of the vulnerabilities imposed by pedLTC, as well as the environmental factors (family,

community, and health, education and social systems) that bolster or diminish the capabilities of young adults with pedLTC to achieve their goals (Panter-Brick & Leckman, 2013; Rutter, 2013). For example, dynamic and coordinated processes of navigating and negotiating for meaningful and available resources generally enhance resilience (Ungar, Brown, Liebenberg, Cheung & Levine, 2008). Further, contextual developmental processes and external supports such as family, community, coordinated health, social and educational programs, and resources may support their resilience and reduce their vulnerability (deCamargo, 2011; Hamdani et al., 2011; Supkoff et al., 2012; Ungar et al.).

Coping and the Appraisal of Stress

Coping is a process oriented and contextual response to manage demands that are perceived to exceed personal resources (Folkman, Lazarus, Dunkel-Schetter, DeLongis & Gruen, 1986). Personal factors play a role in both resiliency and coping; however, it is the unfolding developmental process that is most important determinant of outcomes (Folkman et al.; Panter-Brick & Lickman, 2013). For young adults with pedLTC, the profound impact of their condition and declining health trajectory may determine their capacity to prevent, overcome or improve the personal or external transitional stressors. Process oriented views of coping focus on opportunities for a variety of coping strategies depending on how the stressful event unfolds, or in the case of young adults with pedLTC, may be repeatedly encountered. Coping strategies are neither positive nor negative, thus what is adaptive or unsuccessful in one circumstance may not be in another

(Folkman et al.). When stress is appraised, common coping methods are to alter the situation or accept it, seek more information, and restrain from acting impulsively. Young adults with pedLTC may view the stressors they face in various aspects of transition as harmful or threatening; or challenging, and an opportunity for mastery and success (Folkman et al.) depending on the contextual and environmental resources available to them.

To date, the literature on transitions from pediatric to adult systems demonstrates that the requisite contextual resources in health, education and social systems for navigation and negotiation are lacking (deCamargo, 2011; Doug et al., 2011; Lotstein et al., 2009). Further, securing supportive physical, psychological, social and physical resources may foretell the success of young adults with pedLTC in adapting to adversities, especially those related to the transition from pediatric to adult services (Ungar, Ghazinour & Richter, 2013).

Methods

Research Design

This study is part of a larger descriptive case study (Yin, 2009) that explored 1) the experiences of young adults with pedLTC who have transitioned from pediatric palliative services to adult care; 2) the significant supports and factors that contributed to, or were barriers to, achieving their developmental goals; and 3) the advantages and disadvantages of an online focus group to engage medically fragile participants. Accessing this population with traditional qualitative interview methods, such as focus groups, is difficult because their

numbers are small; they are geographically dispersed; the complexity of their conditions requires many supports, such as care attendants for personal care and respiratory support; equipment, such as power wheel chairs and computer assisted communication devices; and because of divergent mobility and communication abilities related to the impact of their conditions. An innovative method of data collection, a bulletin board focus group (BBFG), which is an asynchronous modification of an online focus group, was used to overcome these barriers. Two university ethical review boards approved this study. This case study was conducted over 12 months in two phases: the first phase consisted of in depth face-to-face interviews, and the second phase consisted of the development, testing and implementation of the BBFG. Details of this process are described elsewhere (Cook, Jack, Siden, Thabane & Browne, 2013).

The Sample

A purposeful sample of youth with pedLTC, aged between 19 and 29 years, with limited expectations to live beyond their first decade of adulthood (29 years), were selected from the graduates of a children's hospice/palliative care program in Western Canada. Their pedLTC diagnoses included Duchennes Muscular Dystrophy (DMD), Spinal Muscular Atrophy, Friedrichs Ataxia, and an undiagnosed neuromuscular condition. From the small sample of these graduates, those meeting the following criteria were eligible: English speaking, cognitively able to participate in their care and decision-making, owning a computer, and having either verbal or typing ability sufficient to participate in face to face

interviews or an online focus group. Most of these young adults require 24-hour attendants for personal, respiratory and feeding support. Their independent function may be limited to minimal movement of their index finger, or a dot on their forehead to control their power wheel chairs, computers and phones. All the participants have lived with their conditions for more than 10 years, and most were diagnosed when they were less than 5 years old.

A purposeful sample of professional consultants including experts in DMD, pediatric palliative care transition, and youth disabilities and sexuality were also selected to participate in this study.

Data Collection

In the first stage of this case study, two young adults with diverse and unique experiences that differed regarding characteristics such as diagnosis, age, gender, family support, opportunities for post-secondary education, and living arrangements, were invited to participate in three face-to-face interviews. These participants were similar in their physical limitations, requiring 24-hour care and possessing minimal movement adequate to control their power wheel chairs, computers and phones. One participant was able to speak directly with me, and the other used a computer-assisted device to type out answers. Interviews were also conducted with an expert in transitional services for young people with neuromuscular conditions, and an expert in the developmental needs of young adults with disabilities.

Face-to-face or telephone interviews with expert consultants (condition specific, young adult sexuality, social support for housing) were conducted to fill in gaps in knowledge arising from the young adult interviews. These participants were recruited through email.

In the second phase of this study, a BBFG was conducted with two groups of four young adults with pedLTC. These participants were selected to achieve variety among their diagnoses, gender, age, and post-transition experiences. The BBFG provided an opportunity to bring together small groups of young adults with pedLTC who otherwise could not be convened. This is the first circumstance of using a BBFG to gather data from this population.

Data analysis

Face-to-face interviews with participants and experts in pedLTC were taped, transcribed and imported into the NVivo 9TM software analysis program. Transcripts from the focus groups were also imported directly from the iTracksTM BBFG discussion board into NVivo 9TM. Conventional and directed content analyses (Hsieh & Shannon, 2005) were used along with a constant comparative analytical process to code the raw data and review emerging patterns, themes, variations and relationships among the data sets (Miles & Huberman, 1994; Patton, 2002). Case study analysis was directed and confined by the theoretical propositions that frame the study (Yin, 2009). Credibility was ensured through use of complementary case study data collection strategies such as face-to-face interviews with young adults with pedLTC and health and social service experts

in young adults with disabilities; online focus groups; content analysis of the literature, program recommendations and professional and jurisdictional policies; and review of the data by pediatric palliative transition experts and participants from the BBFG.

Results: Life Investments

Characteristics of the Participants

Fourteen participants were invited to participate in the study, with 10 consenting to complete an individual or group interview. Two participated in face-to-face interviews and eight young adults participated in the BBFG. Participant characteristics are summarized in Table 16.

Table 16

Participant Characteristics for Face-to-Face Interviews and Bulletin Board Discussion Groups

Conditions	# of Participants (N=10)	Age Range and Mean	Gender
DMD	4	20-28 23	Males = 4
Spinal Muscular Atrophy (SMA)	3	21-26 23	Females = 3
Other: Undiagnosed, Brain Tumor, Friedrichs Ataxia	3	20-22 21	Males = 2 Females = 1

Investing in the Future.

“If we focus on what we can’t do, we’ll never get anywhere.” With the enormous challenges that these young adults face managing their conditions and

transitioning from pediatric to adult systems, it is easy to compile a problems list. However, the participant data is full of examples of persevering and creative strategies that demonstrate their will to transform their vulnerabilities and their beliefs that they have control of their lives, rather than living within the confining framework of life threatening and debilitating conditions. This section will describe the experiences of young adults with pedLTC “getting stuck”, the personal characteristics and strategies that were transformative, and the creative, resilient and persistent journeys they undertook to achieve their personal goals.

Finding Meaning and Belonging

Young adults with pedLTC require an enormous amount of time and energy focused exclusively on their daily personal needs. Activities of daily living, such as getting up and going to bed, showering, getting dressed, eating meals, and doing exercises are considered “work” and are tiring, and require time for rest. In addition, many of these young adults experience chronic and debilitating pain. As a result of the lack of time, energy, and resources in addition to the fatigue and constant pain experienced, engaging in broader activities such as taking courses, working, or creating meaningful pursuits is almost incomprehensible. Creating meaning and purpose, and expressing appreciation, were desired intently and with great diversity among all of the participants. Meaningful activities paralleled young adult developmental goals such as pursuing vocation through post-secondary education or personal interests; engaging in volunteer and faith based

communities; creating a supportive network of friends; and just living an “ordinary” life.

Post-secondary education and vocation. Post-secondary education and vocation were the most time-intensive pursuits of all meaningful activities for the participants. All of the participants were enrolled in or planning to attend post secondary institutions, working, or hoping to find work. While they were hopeful about work, they experienced significant barriers and some discrimination when trying to secure opportunities that will accommodate their disabilities. For example, one participant commented, “A lot of people when they see people in wheelchairs they automatically think that we’re not intelligent or we have a mental condition.” The presence of their support care attendants, and modifications required to access bathroom facilities, were the most substantial obstacles described. Even when employers provided the necessary accommodations, participants had to contend with discrimination or complaints from fellow employees. Many participants were optimistic about opportunities to work online; e.g. as pharmacists and journalists, or in other creative niches. Other participants described finding vocation in activities such as writing a book about their experiences or setting up their own small business making T-shirts.

Awareness of the fragility of their time remaining contributed to agonizing decisions about pursuing-time intensive goals such as post-secondary education. Unlike other young adults, those with pedLTC do not have their “life ahead of them” when they complete post-secondary education. Taking into

account the physical limitations imposed by their condition, and the time required for treatments and personal care, an undergraduate degree will take them longer than four years. For some, pursuing post-secondary education conflicted with finding time enough to pursue pleasurable activities such as being with friends. This struggle was more poignant for those who were already experiencing symptoms of declining health, compared with those who anticipated, but were not experiencing, decreasing function. One participant described her inner battle:

Of course I do want to graduate and get a job and stuff, but if I always limit myself to like the few years that I have left, I won't be able to do that because I'll just be living in a time frame, and just wanting to spend my time doing fun activities with my parents and my friends, instead of actually pursuing my life goals.

For those who had not experienced significant changes in their health status, hopes to pursue and achieve their dreams were less impacted by the reality of decreasing time.

Volunteerism. Finding meaning in higher purposes such as faith based community groups and supporting charities was widespread. For example, using a computer-assisted program that was operated by a “button” on her forehead, one participant made and sold a range of cards, giving the proceeds to charity. Unable to verbally articulate the meaning of living with her debilitating condition, another participant wrote spiritual songs, poems and testimonials that were read or performed to groups by her close friends. Others volunteered at camps for

disabled children, offering back or improving upon the mentorship and support they received growing up. Finding meaningful volunteer activities sometimes was challenging and not always successful. For example, one participant found that she was not a welcome visitor for shut-in seniors because of the concern that her wheelchair might scratch their furniture. And another, ironically while volunteering for the cancer society, was diagnosed with cancer. Despite these distressing complications, they persevered to find alternate volunteer activities.

Online gaming. Online gaming was a meaningful activity for about two-thirds of the participants. They were able to establish a wide range of friendships beyond their disabled community, and to focus on broader goals not related to their conditions. One participant met her husband, now her most meaningful relationship, online. Online friendships are a wonderful outcome of technology, and are reflective of changing societal patterns of friendship. Online games provided opportunities to overcome impossible hurdles and obstacles that limit their physical worlds but not their virtual worlds. After a day of frustration with service providers, or not feeling valued or recognized because of their limitations, the virtual world provided a welcome solace.

While online gaming facilitated an escape from isolation for some, it did perpetuate isolation, and for others replaced socializing almost entirely. Further, online games appeared to have a numbing effect, insulating them from the reality of their isolation and vulnerabilities. Finally, declining function impacts their ongoing access to online games. Depending on computer technology, games that

require arm strength, dexterity or stamina may become beyond their ability.

Participants were hopeful that technological advances would continue to support their needs.

Socializing. Sociability with friends, either on line or in person, and playing team sports, were rated as important endeavours. Isolation—whether by choice or due to circumstances such as being housebound or losing communication abilities—was often associated with feeling stuck. For some participants, being recognized as unique and noticeable was energizing and empowering, whereas others struggled with feeling different and not accepted outside of disability groups. Believing in oneself, and not the opinions of others, was a successful strategy for fostering resilience in the face of negative remarks and attitudes, and active social networks. For others, social connectedness was difficult to achieve because of limited speech capacity requiring technological support to communicate, limited age-appropriate community programs, and paradoxically, the impact of their communication equipment in creating a screen between them and other people. For those with an interest in sports, power wheel chair sports teams offered regular opportunities for socialization with other young adults with similar conditions, a place to belong, camaraderie, and meaningful purpose in developing personal and team building skills. For some, wheelchair sports teams also facilitated travel to international events. These opportunities were not available to those with a preference for the arts.

Intimacy. The most challenging conversation in which to engage with the participants concerned intimate relationships. An expert in sexuality for disabled confirmed that the disabled generally don't have the vocabulary to discuss sexuality because they miss out on normative experiences in school, and on being invited to gatherings of young people where most adolescent intimacy takes place. If they are able to attend these events, parents may need to accompany them as care attendants. Further, a gender discrepancy favors young men's sexuality being supported by parents and care attendants, who may help them engage in sexual experiences through sex workers and masturbation. Young disabled women are expected to be sexually passive and silent, while the dominant cultural narrative creates an atmosphere of acceptance in which young men may seek sexual experiences before they die (Liddiard, 2013).

Those who participated in this conversation struggled against a societal view that stereotypes disabled people as being asexual. They emphasized that physical limitations become irrelevant when you are known and loved for who you are. One married participant felt devalued and one-dimensional when others assumed that her husband was her care attendant, brother or friend. The incredulous responses she encountered confirmed society's normative beliefs that people with disabilities are asexual.

Being Hopeful and Realistically Optimistic

Hopefulness for the future was universal among the participants, matching young adult developmental goals of going to school, having work, living

independently and being involved in intimate relationships. The BBFG participants were asked to upload images, pictures, or songs that represented the future for them. A computer keyboard with a “future” key symbolized excitement about a future with new technologies guaranteeing self-expression and social connectedness, even with declining function. The phrase “accept what you can’t change and change what you can’t accept” embodied a realistic outlook on both their possibilities and limitations. A picture of the participant’s head photo-shopped onto a competitive swimmer’s body exemplified hope for transformational change through a cure; a serene picture of a pathway bordered by autumn trees and peaceful water epitomized hope, that while the twists and turns on the path are unknown, the journey will be beautiful.

For some, constraining the future in a short time frame facilitated realistic hope and dispelled ambivalence and despair about not having a future. Typical 5 and 10-year plans felt unrealistic from the perspective of living with progressive disabilities. Six-month and yearly goals were more credible and allowed room to hold two competing realities: while their future is uncertain, achieving some of their dreams may be realistic. For some, hopefulness extended beyond wishing for their conditions to remain stable; they were hopeful for improvements with new treatments, technologies, or a cure. For others, even though unknown, the future is “bigger and better” with opportunities for university, vocation, and building lasting relationships.

While their conditions and anticipated end of life trajectory varied, optimism was unwavering, even for those experiencing declining wellness and function. As one participant stated, “So, like I do notice changes, but I try to ignore them and live my life just ignoring my disability and trying to think positively.” In the context of living with profoundly debilitating conditions, “ignore” is interpreted as a strategy to keep their condition in its place, and not let it take center stage. As this participant describes, without this resilient strategy, life would be bound by limitations and treatment regimens:

I’ve chosen to live in the moment instead of letting my disability control my life . . . but I don’t know if it will work, in like five years down the road, but I guess that’s why I’m so adamant about what I want to do. So I study, or like Facebook, instead of spending my time, like all in medical procedures.

This resilience and optimism creates tensions with parents, who are more focused on the requirements of the disease, than on the young adult’s pursuit of their goals. Chatting on Facebook and hanging out with friends, instead of resting and doing physiotherapy exercises, are examples of tensions that arise between young adults and their parents. The young adults value living in the present, whereas the parents value longevity and maintaining treatment regimens.

Managing Expectations for the Future

In the BBFG groups, the participants were asked to rank their expectations of their condition remaining the same, improving, or declining in the next two and five years, as outlined in Table 17.

Table 17

Ranking of Future Expectations

Expectations of condition	In 2 years	In 5 years
Improve	25%	12.5%
Stay the same	75%	62.5%
Decline	-	25%

While two participants indicated that they expected a decline in their condition in the next five years, their comments remained hopeful and optimistic, with an undercurrent that demonstrated realism and an understanding of their disease progression. One participant with DMD commented to the BBFG group: “I know that doctors are probably not very confident with my statement, and I am realistic, but I want to stay on the positive side.” For young adults with pedLTC, thinking beyond a few years was difficult. One participant captures the ambivalence, and perhaps reticence, to look ahead into an uncertain future that is likely fraught with changes: “Truth is, I don’t know, but either way, what is is, and I plan to keep a positive attitude.”

Strategies to manage the future included being adaptable, being prepared, and mentorship. For some, living with a progressive disease has created adaptability and “living with life as it comes” knowing that things are always changing. On the other end of the spectrum, being prepared for an uncertain

future became a personal coping strategy. For example, one participant discussed this strategy related to preparing early for exams in case she was tired or unwell close to the exam date. Mentorship from older friends, and likewise mentoring teens with the same condition, were suggested as successful strategies to take control of their future.

For participants with neuromuscular diseases, being realistic about their future was imperative because a respiratory crisis or change in their ability to breathe independently was inevitable. For these young adults, realism required embracing a potentially fearful and uncertain future of deciding whether they want to live longer with a ventilator, or live without mechanical ventilation and die sooner without respiratory support. Making a decision prior to this future event was essential because at the moment of crisis, reflective decision making about the implications of living with a ventilator is not possible.

Empowerment: Getting What You Need

Much of the transition literature refers to the transition phase in terms of disempowerment: it is like falling off a cliff (vanStaa, Jedeloo, van Meeteren & Latour, 2011), because the current health, education and social systems do not support their needs. For young adults with pedLTC in this study, the loss of comprehensive pediatric services was profound, and especially difficult if they lost support from their family and community. System barriers did limit and sometimes impede their efforts to live their young adult lives to their fullest, even while their health condition was stable. For some, maintaining their personal

safety required a battle for safe living conditions. In addition to system barriers, family also created barriers to these young adults gaining independence that required them to “take charge” and gain control of their destiny. The next section describes the strategies used by these participants to get what they needed from systems, programs and personnel to pursue their goals.

Fight and advocate. Frustrations with the system provided purpose and opportunities for meaningful involvement. Rather than accept the limited resources offered in the adult systems, some participants described “fighting” for everything, never taking no for an answer, and never giving up. The “fight” became a powerful response to unresponsive systems, services and personnel. However, for those that predominantly used a “fight, fight, fight” metaphor to describe how they get what they need, they did not have a repertoire of other strategies or a “tool box” to use when “fighting” did not work. While successful in some circumstances, it was maladaptive in others. For some, their condition reduced their speaking capacity, and they had to rely on email rather than face-to-face or telephone conversations to “fight.” They described “hounding with email” as another “fighting” strategy.

For others, an advocacy style was employed to get what they needed from the system. Advocacy was described as neither fighting nor being passive, but finding ways to collaborate with the people working where system barriers exist. For example, using “win-win” strategies that matched the values and mission of the target system or institution with their needs was successful. Those who had

success in negotiating for services or facilities used task-oriented tactics requiring incremental and purposeful action. Other important advocacy skills included learning how to negotiate professionally, non-aggressively, and how to “pull on heart strings.” Negotiations were more successful when their village of significant community members, such as pediatric physicians and physiotherapists, provided letters of support. Among those without a larger community of support from family and pediatric providers, many felt stuck and overwhelmed by system barriers that thwarted or delayed their plans for post-secondary education and vocation. Finally, others experienced more successful negotiations when their communications were persistent, and expressed gratefulness and thankfulness for the services they were receiving.

These persuasive, creative and engaging strategies with system service providers were not always successful. For some, maintaining personal control and safety became a battle for their life. For example, in an extended care facility, when a participant’s call bell was not answered and he required suctioning, he “Face booked” his friends and asked them to call the nurse’s desk at the extended care facility to tell them that he needed help right away. On another occasion, his pediatric support person found him in his room drowning in his secretions with the ventilator and emergency call bells ringing. Being underfunded for safe ventilator support complicates accessing independent living. The fear of being unable to get help and support quickly for worsening fluid secretions in their lungs or ventilator problems either ignites a powerful fight for a higher level of

support, or acquiescing to the status quo and continuing to live with family who provide some of the needed missing support hours.

Taking charge. For young adults with pedLTC, taking charge of their destiny and learning to be independent was described as the most significant hurdle to overcome. While they are dependent on help for all physical activities of daily living, cognitively, they are very independent. For young adults with pedLTC, independence is not related to reliance on others for help. Paradoxically, support provides a level of independence that cannot be realized without it.

Parents who did not treat their young adult with pedLTC any differently than the other siblings encouraged autonomy, and fostered independence (Stam, Hartman, Deurloo, Groothoff & Grootenhuis, 2006). For example, one participant described his parents' strategy this way:

If you want to go somewhere, "go take the bus." They were never afraid to send me on my way and to be independent from a very young age. Once you learn that you can in fact do things on your own it develops your confidence and you begin to take charge of your life.

Further, being unable to function independently is not a deterrent to being independent. Not being able to ring the stop button on the bus, and press elevator or crosswalk buttons, encourages learning how to ask for help. From the young adults' perspective, taking charge requires believing in your strength to achieve more than others believe.

Taking charge was diminished by a reciprocating dependency pattern between the parents who need and want to help, and the young adults who learn to believe they are both disabled and unable. As one participant describes, “For me it’s been difficult because my parents could not let go of me as their disabled daughter, and let me grow up and carry on with my own life.” After two decades of caregiving, relinquishing both caretaking and decision making roles for their vulnerable young adult with pedLTC is nearly incomprehensible. Further, many parents did not expect their children with pedLTC to live into adulthood. However, maintaining dependence makes it difficult for these young adults to have dreams and achieve them.

Ambivalence and a propensity to anticipate that some tasks, such as ordering equipment, and hiring, firing, and organizing the schedule of their care attendants, will be too overwhelming, also hindered taking charge. Looking into the future, other participants anticipated their parents’ decline, and the necessity of taking charge of managing their day-to-day activities and advanced directive decisions sooner, rather than later.

Accept Your Life

Accepting your circumstances, believing in yourself and not what others think about you, and achieving more than others believe you can, were recurring themes for these young adults, regardless of the specifics of their condition. These supportive maxims were foundational to their resilient and transformative experiences. For example, in a discussion about how their disability and

equipment can get in the way of people really knowing who they are, they were invited to upload onto the BBFG an image or song to represent what they wished people would see instead of them in their chair. Two of the respondents posted a picture of their power wheelchair with these comments: “People will only let your wheelchair define you if that’s all you see too! This is my life and I love every minute of it.” The other participant responded, “it’s funny you say that, because that’s how I’ve always felt too . . . I liked being the girl in my wheelchair, because everybody knew who I was :-)”.

For young adults with pedLTC, “accepting your life,” means more than life; it necessitates accepting declining health, function, and death. Young adults with pedLTC are forging into new territory with few mentors to guide decisions that will impact both their longevity and quality of life. Their most difficult trailblazing experience is the developmental mismatch of preparing for their death at a time of life that is wired for a fulsome life. As one participant comments: “No one likes to talk or even think of end of life at such an early age, but for many of us in similar situations, it is a very real thing.” Transition preparation is focused on getting on with their lives: independent living, education, sustainable funding sources, and forging new relationships with system and program providers. Further, many of these young adults have “cheated death,” either by living well beyond expectations, or surviving medical crises, and may expect to have more “cheating” opportunities.

When asked if they had established advanced directives for their care if

they were unable to communicate their preferences, 33% reported yes, 22% reported no, and 45% did not know. Of those who did have advanced directives in place, they were specific to resuscitation, and not to other life sustaining interventions like a tracheostomy for ventilation, or a feeding tube.

While they acknowledge their conditions are life limiting, discussing their thoughts about their own end of life is difficult. The face-to-face interviews provided more opportunity than the BBFG to gently probe when they were reticent to speak about death. Interestingly, the following factors did not contribute to their ease discussing death: having a religious faith, complexity of their condition, relative age within the group, knowing someone else who died with their condition, and length of time living with their condition. Of those who were willing to speak about their end of life trajectory, independence and the ability to communicate were the key criteria that defined their quality of life, and would influence their advance directive choices. For those who did not have a tracheostomy, 24-hour attendant support required for ventilation was associated with losing independence. For those already ventilated, 24-hour support did not compromise their sense of independence. These young adults were willing to risk spending short periods of time alone, aware that they could die if they had mechanical problems, or a build up of secretions they were unable to clear without suctioning. Prior to ventilation, decision-making was challenging, whereas after the tracheostomy, it was not viewed as a monumental decision. Being prepared for this decision was one of the most important tasks of

adolescence described by participants. The quality of communication preferred varied from technological support for typing, to eye and facial movements to convey preferences and feelings.

Discussion

Many of these young adults with pedLTC demonstrate a striking capacity to transform the personal circumstances imposed by their pedLTC and the contextual barriers they experience transitioning from pediatric to adult services. Coping and resilience theories (Folkman et al., 1986; Rutter, 2013) provide a helpful framework to understand contextual variables, such as systems, family and community supports (Ager, 2013), and opportunities for successful coping with stressful challenges (Rutter) to support resilient and transformative outcomes. These components of coping and resiliency will be examined in the context of young adults with pedLTC.

Success was enhanced when requisite resources were available, coordinated, navigable, and responsive to new and unique requests (Ungar et al., 2008). Being unable to make transformative changes occurred when personal characteristics, or contextual and environmental supports, were lacking (Supkoff et al., 2012). Further, transitioning from pediatric to adult services, and adolescence to young adulthood with pedLTC, was not an “either/or” experience of resilient transformations, or being thwarted, but rather, an iterative process.

Living a Paradox

Life for young adults with pedLTC is peppered with paradoxes. For example, accepting that independence requires being dependent on caregivers, power wheelchairs and other equipment, provides more opportunities for self-fulfillment and autonomous choices. Developmentally, opportunities for achieving personal goals, independence and vocational and educational pursuits diminish with declining health status. In contrast with supportive pediatric services that extend and improve their quality of life, uncoordinated adult services and reductions in services and programs result in increased morbidity, more visits to hospitals, and rising health costs (Melies et al., 2000; Prestige et al., 2012). Further, these young adults face a barrier to palliative services as they come closer to the end of their life. Living within paradoxes, and experiencing declining health, function and system resources, requires continually reconstructing a new normal on their journey forward (Walsh, 2012).

Contrasting Outcomes

For some, long-term living with their condition with environmentally supportive pediatric and palliative programs and systems may enhance resiliency (Panter-Brick & Leckman, 2013; Walsh, 2012). Adaptability to the changes imposed by their progressive conditions, achieving academic success despite missing traditional classroom time, and accepting living experiences that are widely divergent from other children their age, may support resilience for some, while others are disempowered by the burdens of their condition (Freyer, 2004; Knafl et al., 2006). Many were able to keep illness in its place through their

ability to adapt, adjust and transform their experiences (Knafl et al., 2006; Walsh, 2012). Some participants had fewer coping strategies and got “stuck” more easily. It appears that unrelenting stress from lack of family support, inability to engage in meaningful activities, or a supportive social network may sculpt the brain into reliance on one coping pattern, such as “fight, fight, fight” (Panter-Brick & Leckman). When a system support was lacking, some participants were able to transform barriers by engaging their community of support (Folkman et al., 1986).

Appraisal of stressors. The separation from pediatric services at adulthood is challenging, but for some, a positive appraisal of their ability to overcome adversity, and support from other sources such as pediatric professionals and community members, enhanced their adaptability and negotiation strategies (Calhoun & Tedeschi, 2006; Folkman et al., 1986). Differing appraisals of and reactions to the impact of stress are determined by perceptions of harm or loss, threat, and challenge (Lazarus & Folkman, 1984). For many of the young adults in this study, transition threatens their young adult ambitions, diminishes their belief that the health, education and social systems will support them, and challenges them to make changes that do not align with their condition-specific needs.

For young adults with pedLTC, their appraisals of the threats, harms/ losses, and challenges experienced by their condition, overlap with and become magnified by appraisals of their transition experience. Further, the meaning they attribute to living with their condition, and their transitional difficulties, will

shape their behavioural and emotional responses (Lazarus & Folkman, 1984). The same event can produce different reactions in people experiencing the same threat. Table 18 demonstrates differing responses between individuals to the stress of losing attendant support hours at transition. For some, the change in attendant support may be perceived as a threat to achieving post-secondary goals, loss of independence, and a challenge to their ability to socialize. Others may perceive this as a loss of comfort, harm to maintaining functionality without support for exercise, and a threat to maintaining their health status.

Table 18

Variations in Appraisal of Stress (loss of attendant support hours) Related to pedLTC Transition-Specific Stressors.

	Goals: Post Secondary, Vocation, etc.	Independence	Health Status	Function	Comfort	Socialization
Threat	Person x		Person y			
Harm				Person y		
Loss		Person x			Person y	
Challenge						Person x

This table demonstrates appraisal responses to one change in one moment of time. However, young adults with pedLTC will be faced with a myriad of these types of stressors simultaneously. In addition to appraisals of stress varying among individuals, appraisal and the meaning attached to them will also vary across time and within individuals. The level of threat, harm, loss and challenge

they experience will be determined by their perception of their condition being stable or declining. Further, reappraisal of the stressors based on new information and experiences will be continuous and ongoing (Lazarus and Folkman).

Skilled and astute clinicians are required to assist these young adults to unravel and reframe the complexity of the ongoing challenges imposed by transition, and their own declining function. To be supportive to these young adults facing complex, challenging, and changing stressors, it is important for clinicians to appreciate that there are multiple interacting factors influencing their response. Some of these factors will include their perception of specific changes as stressful or a challenge and their perception of the status of their condition; previous successes coping with changes; support from family, friends and their community; and other personal and environmental factors. The interplay of these factors will determine their response and whether they can harness positive strategies, or feel overwhelmed, retreat and become passive. The following types of questions will be helpful for clinicians to support these young adults unravel, reframe, and appreciate the complexities they face:

1. Will you feel threatened, harmed or challenged by *fewer attendant support hours*?
2. What will you lose with *fewer attendant support hours*?
3. Which of the losses and changes you have described is the most important loss to you?
4. What will these losses and changes mean to you?

Naturalistic Opportunities

Understanding the environmental, contextual and system deficits these young adults face is important in determining for whom, and when and how resources truly matter (Panter-Brick & Leckman, 2013). Resilience oriented approaches seek out naturalistic “islands of strength and resources” (Walsh, 2012, p. 174). For those who were fortunate, having a village of family, friends, community leaders, coaches, teachers, and health care professionals contributed to their resilience lifelines. Others got stuck and were unable to effect the changes they desired. While more opportunities are available in urban areas for independent living, access to vocational and educational opportunities, transportation, socializing with friends with similar conditions, and pursuing sports, online connectivity with mentors in the creative arts could provide the momentum and inspiration for young adults with pedLTC to achieve their goals.

Parents are important facilitators and advocates for normalizing developmental growth. It is common for youth with chronic health conditions to acquiesce to their parents’ taking charge of their health experiences (Rosen et al., 2003; van Staa et al., 2011), and because of limited opportunities for autonomy and social connections, these young adults will have achieved fewer young adult developmental milestones than their peers. For this young adult population, conflicts with parents arose when their struggle for independence clashed with the confines and management of their illness, and established parental management of their condition (Stam, Hartman, Deurloo, Groothoff & Grootenhuis, 2006; Wein,

Pery & Zer, 2010). Compliance with medications, and health recommendations and procedures such as exercises and respiratory care are probable battlegrounds, in which the young adults fight to assert their independence and seek control developmentally later in young adulthood, rather than adolescence (Stam et al.).

Youth programs that nurture naturalistic opportunities to participate in activities that develop life skills, and provide sustained and caring adult-youth relationships, also improve adult outcomes (Maslow & Chung, 2013). For youth with chronic health conditions, and even more so for those with pedLTC, community integration and opportunities for support may be missed because of absences from school due to illness and logistical constraints of programs to accommodate youth with complex conditions. The Chronic Illness Peer Support (ChIPS) program's objectives mirror many of the resilient and transformative attributes of the young adults with pedLTC, providing a template to design programs that: 1) promote coping techniques to influence social environments, understand personal stressors, learn to take on alternate perspectives, make positive attitudinal changes and enlarge what is perceived as "normal," 2) reduce isolation through group identity and extending help to others, and 3) provide leadership opportunities (Olsson, Boyce, Toumbourou & Sawyer, 2005). Further, specific attention to declining wellness, increasing symptoms and periodic health crises are essential for young adults with pedLTC. Considerable research, activism, and storytelling are required to move towards positive sexual examples and cultures for disabled people (Liddiard, 2012).

Uncertain Futures

Conversations initiated in pediatric palliative care program transition programs that discuss future oriented strategies to access independent living, vocational and educational opportunities; health decisions affecting independence, communication and quality of life; intimacy; and advanced directives for end of life, will have more resonance in the young adult years as they face the reality of their declining health, observe friends die from similar conditions, and are able to view these issues through a more mature developmental lens (Fryer, 2004; Rosen, Blum, Britto, Sawyer & Siegel, 2003). Framing future thinking along a continuum of avoidance and engagement (Fletcher Johnson, 2012) aptly describes the experiences of these young adults who are aware of the importance of both living in the moment and making plans for their future. Supportive peer and professional relationships are required for a balanced consideration of living in the moment and peeling away layers of defense regarding an uncertain future (Shaw & DeLaet, 2010; Wein et al., 2010). Young adult hospice programs can offer these relationships and provide this social connectivity through professionals, peers and volunteers, and access to these conversations through online forums and group meetings.

Study Strengths and Limitations

While this is a small study and the results are not generalizable, it is among the first research studies to document transition experiences from the perspective of young adults with pedLTC. Further, this study expanded the

concept of stress appraisal beyond one appraisal. Additionally, data were collected using an innovative method, the BBFG, to reach a medically fragile population. While there were instances of limited follow-through by some participants on the BBFG discussion threads, the resulting data were rich and creative. Further research is needed to understand the interconnectedness of personal characteristics and environmental supports, and how to empower these resources to enhance resilience and developmental life achievements, such as vocation and education for young adults with pedLTC. Further research is also required to understand how to support intimacy, and end of life decisions and care.

Conclusions

Documenting the experiences of young adults with pedLTC overcoming personal and environmental odds to achieve their life goals is inspirational. Their experiences demonstrate their capacity to maintain their vitality in the present, be hopeful for the future and persevere to invest in their lives. Learning from the personal and contextual factors that enhanced or inhibited resilience and coping strategies will provide direction for the kinds of supports and resources most beneficial in the transition from pediatric to adult services. Efforts initiated in pediatric care need to be followed through into young adulthood, with specific advocacy training to foster confidence, create mentor relationships, and maintain social connectivity into adulthood (Maslow & Chung, 2013; Olsson et al., 2005). System resources are imperative to provide the necessary resources such as counselling and mental health services, support for educational and vocational

opportunities, development of community programs, and socioeconomic improvement. Connectedness across levels of practice and policy, and education, health and social systems is important to create synergistic effects of intervention (Ager, 2013). Young adult hospice programs can provide the resilience lifelines required to counter the risks associated with declining health, and decreasing support from health, social and educational services to support young adults with pedLTC achieve their dreams and enhance their quality of life.

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CHAPTER 6

REFLECTIONS AND CONCLUSIONS

REFLECTIONS AND CONCLUSIONS

*I don't feel like I fit into the world
People don't see me because I am in a wheelchair
Only the people who know me see me as a person
I am the same as everyone else my age*

(Used with permission)

These lines, written by one of the participants, introduced this dissertation and the experience of a young adult living with a pedLTC. Throughout this study, other participants described similar young adult desires: to be known, to be seen as a person, to be acknowledged for competency and ability, to be given a chance, to be “normal,” to have hope, to write a book, to have intimacy, to go to school, to live independently, to get a job, to run a business, and to travel. These are not unusual aspirations, but as this study describes, they are monumental achievements for young adults with pedLTC.

The purposes of this study were to lay a foundational overview of the transition experiences of the first generation of young adults with pedLTC to live into adulthood; to understand the opportunities and challenges imposed by the current health, education and social service systems; and to develop, implement and evaluate a novel research strategy for reaching medically fragile young adults with pedLTC. This study offers insights into the experiences of this first generation of young adults with pedLTC to live into adulthood, and the potential for online modalities to enhance research and social connectivity with these young adults.

The results of this study covered several domains. First, the impact of health, education and social systems inadequacies, insufficient support, and lack of integration were found to delay or thwart young adults with pedLTC from achieving their goals. Second, despite these system inadequacies and condition-imposed vulnerabilities, many of the young adults were able to mobilize a supportive community and persevere and persist against barriers to transform their experiences. Third, this study uncovered multiple paradoxes for these young adults, such as independence requiring dependence; hopeful futures imposed on progressively declining conditions; young adult aspirations colliding with end of life; supportive services intended to enhance and prolong their lives removed; vulnerability transforming into resiliency; and becoming disconnected from palliative and hospice services as end of life approaches. Fourth, while uncertain futures were clinically expected, hopefulness and realistic optimism were evident. Fifth, the opportunities and benefits of online strategies with medically fragile populations were demonstrated. Finally, this study revisited well-known theories of vulnerability, coping and resilience, and uncertainty, to appreciate these young adults experiences and outcomes while navigating through transitional and developmental incidents within a limited life trajectory.

This chapter will review the findings of this study and offer further interpretations of how these results address the research questions; provide a conceptual representation of the interconnectivity and relatedness of significant

theories and topics; and suggest strategies for practice and policy, and implications for future research.

System Impact

The current health, education and social systems do not fully support the transition of youth from pediatric to adult services. When faced with the inevitable gaps in the systems and lack of coordinated services among the health, educational and social sectors, the participants employed various thoughtful and novel strategies to get what they needed to move forward with their lives. When they came to an impasse in the education and social support systems, some participants were successful navigating and negotiating for what they needed when they encountered barriers in the health care system. Their successful strategies included: thorough research to determine all the available services and how to access them; using their “village” for support and advocacy; being persistent, positive and creative; and never taking no for an answer. However, in other circumstances, the systems were too formidable to impact, and they were stalled or unable to move forward.

The young adults suggested the following recommendations to improve system inadequacies: 1) knowledgeable providers in health, education and social service sectors and better communication among these providers, 2) coordinated and accessible services within and across the sectors, 3) being respected and valued, and 4) opportunities for independence. Cross system linkages are necessary to facilitate timely and efficient support and provision of services

between the pediatric and adult health, education and social systems to support and enhance these young adults' limited remaining years. Centralized leadership of health, education and social services is necessary to streamline and coordinate programs and services.

Another detrimental system impact is the labeling of young adults with pedLTC as disabled, not palliative; diminishing the essence and experience of their palliative and progressively life limiting condition. In terms of medical management, health care providers may not be attentive to the significance of changing and declining symptoms and unaware that anticipatory guidance and planning are necessary to prevent ensuing medical crises (S. Poitras, personal communication). Equally important, if health care providers only attend to the physical vulnerabilities of young adults with pedLTC, the opportunity is missed to strengthen and leverage their courage and persevering investment in meaningful opportunities.

A Continuum of Transition Experiences: Transformational and Getting Stuck

Transition is defined as a change, conversion, evolution, shift, alteration or modification (Free Online Dictionary), implying that most transitions have a positive outcome. The young adults in this study shared a common transition journey, experiencing multiple negative and frustrating barriers, mostly from health, education and social service system barriers and inadequacies, which prevented them from engaging in their hoped for endeavours. However, for some

of the young adults, the barriers were overcome, and their transition path was transformed. Transformation is a specific type of change that invokes being strengthened rather than weakened (Bussey & Vise, 2007). When barriers are insurmountable, transformative experiences do not occur; the young adults get stuck and are unable to move forward.

Surprisingly, a mathematical example illustrates the iterative process between getting stuck and having transformative experiences. Figure 5 provides a mathematical representation of opportunities for transformative experiences compared to “getting stuck.” The young adult’s health and illness status and personal well being are represented by the y axis, and external supports such as health, education and social services are represented by the x axis.

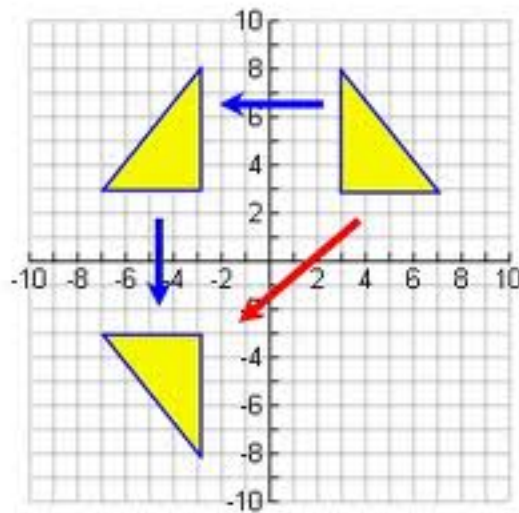


Figure 5. A Mathematical Representation of Transformation and “Getting Stuck”. (Adapted from math.wikia.com). This figure illustrates transformation and getting

stuck in relation to personal resources, environmental support, and impact of health condition.

Prior to transition, if their health condition is stable and pediatric resources are sufficient, they were likely in the top right corner (+ve/+ve). At transition, if their health/illness trajectory and well being remains stable (+ve), but the system services and resources are diminished (-ve), then they shift from the +ve/+ve quadrant to a +ve/-ve quadrant (top left hand corner). The young adults in this study reported that their health status was stable, or changing with small, manageable declines. In this +ve/-ve quadrant, some were empowered to “fight” and transform their situation to get what they needed. Others appeared to be accommodating to declining resources (while support from family was available), or after living with less support than they needed to achieve their goals, were preparing to engage with the system and fight.

If their health/illness trajectory declines (-ve) along with already diminished supports and services (-ve), they shift to a -ve/-ve quadrant (lower left hand corner). This was the experience of one of the participants who had experienced several medical crises, was living in an institution with a nominal community of support, and wanted to live independently. Without the dedicated support of a pediatric provider and friend, he would be remaining in the institution today with little personal agency, depressed, and socially disconnected.

Some of these young adults will experience declining health before difficult transition experiences can become transformative. All have limited

energy, time, and capacity to devote to overcoming the barriers. Others may decide that the goal is not worth the effort and choose other options. Given that the health trajectory of these young adults will decline, current system resources and supports are inadequate, and family support will decline (as their parents become older and siblings may live far away or be consumed by their own responsibilities), many young adults with pedLTC may decline into the double negative quadrant with limited personal agency and supportive resources to enable them to direct their life and end-of-life course. Therefore, it is imperative to ensure that sufficient resources and services are available to support their ability to make significant choices about their life direction within the confines of their condition.

While observing, listening and analyzing the data, I became curious about the ability of some of the young adults to navigate and negotiate around ongoing transition obstacles with a sense of purpose and meaning, while others got stuck and were unable to move forward. What factors facilitated these young adults' ability to be transformative instead of getting stuck? And, what kinds of resources and supports will enhance resilience, decrease vulnerability, and be most helpful for these young adults with pedLTC so they might navigate and negotiate the barriers inherent in the transition process?

The cognitive appraisal process described in “Life Investments” illuminates the differing outcomes and the young people's personal perceptions of their successes based on the meaning they attribute to living with a pedLTC, their

declining health status, and the transition experience. Continuous and ongoing appraisal will determine whether the changes they are experiencing are perceived as a threat, harm or loss, or a challenge (Lazarus & Folkman, 1984).

Vulnerability and Resilience

Young adults with pedLTC experience factors that exponentially increase their vulnerability such as: changing and declining health; profound physical disabilities; debilitating pain; dependence on care providers and equipment for daily living; feeling isolated; the psychological burden of their life threatening condition for themselves and their family; uncertainty about dreaming or achieving goals; and navigating and negotiating new systems (Meleis, Sawyer, Im, Hilfinger & Schumacher, 2000; Simpson & Kettle, 2008; Suris, Michaud & Vine, 2004). However, their perceived vulnerability will be determined by their appraisal of their coping resources, and their perception of imposed transitional and health trajectory changes as a challenge versus a harm, loss or threat (Lazarus & Folkman, 1984).

Another conceptualization of vulnerability is associated with weakness, flaws, frailty or powerlessness. However, the experiences of the young adults in this study demonstrate that vulnerability can be fuel for change and ignite an enormous capacity for resilience and mastery. Recent re-evaluation of vulnerability is revealing that it is not born from weakness, but from courage, and a capacity for emotional risk that can move beyond fear. The power of being vulnerable lies in being comfortable with uncertainty, and being willing to invest

in opportunities not guaranteed to succeed (Brown, 2010). For some of the young adults in this study, the interdependence of vulnerability and resilience spawned courageous and creative endeavours.

Figure 6 provides a visual representation of the inter-connectivity of the participants' experience of the following components of the transition process: appraisal of threat, living with uncertain system support, declining health function, and colliding developmental and health trajectories applied to the theories of vulnerability and resilience. This diagram is not meant to be a comprehensive explanation of transition experiences for all young adults with pedLTC, but rather a visual description of the concurrent and multiple experiences of the participants in this study in relation to the relevant literature discussed. In my observations, the applicability of all these theories to the experiences of young adults with pedLTC was evident. No one theory adequately encapsulates the experiences of these young adults adapting to uncertainty and living multiple simultaneous paradoxes.

The Vulnerability Triangle is adapted from Rogers (1997 in Markle-Reid, 2002), and the Desired Developmental versus Health Trajectory is adapted from Wolfe (2004). The remainder of the diagram is formulated from a synthesis of the study data applied to theories of vulnerability and resilience.

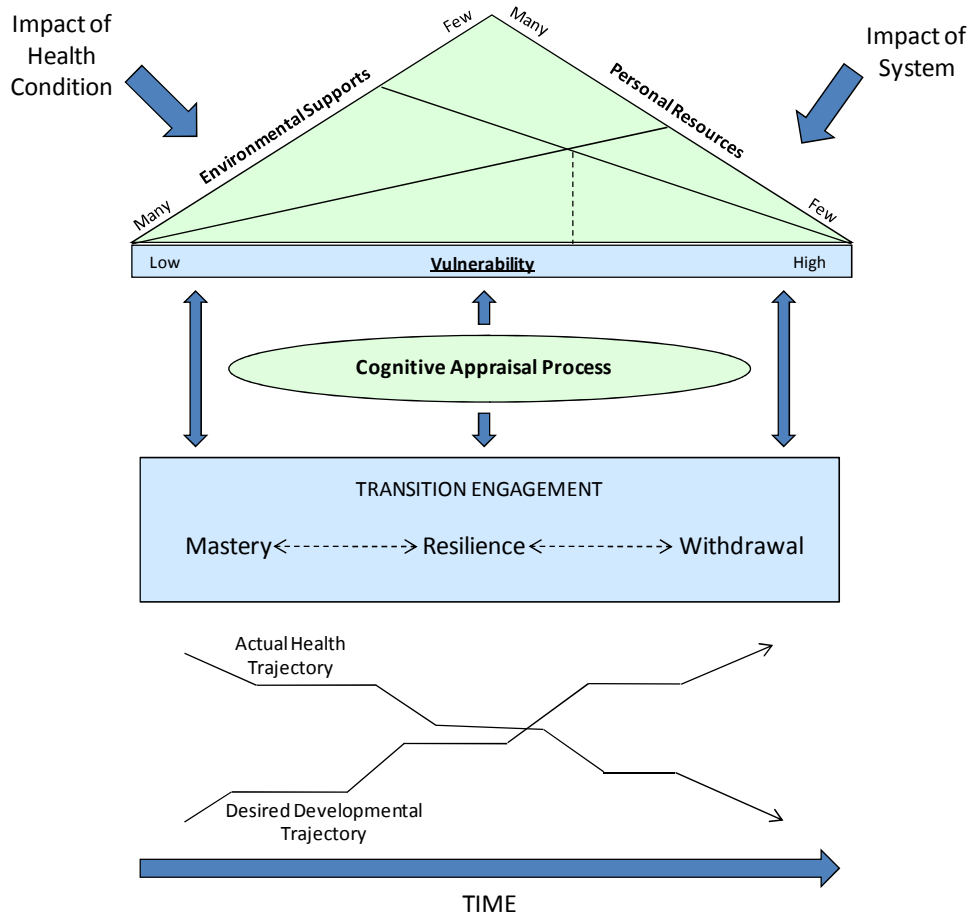


Figure 6. Representation of the Inter-relatedness of Vulnerability, Resilience, Cognitive Appraisal, and Developmental and Health Trajectories.

This diagram demonstrates that for these participants, the uncertainty and variability of their health condition and availability of system resources impact their degree of vulnerability, engagement in the transition process and resilience. As described in Chapter 1, Rogers’ (1997) vulnerability model demonstrates that even if personal resources hold constant, changes in environmental supports alter vulnerability.

To estimate an individual's degree of vulnerability, one would locate the person's level of environmental supports on one side of the triangle and draw a straight line from this point to the opposite apex. The same would be done for the person's personal resources. At the point where the two lines intersect, a line would be dropped perpendicular to the base to intersect the vulnerability continuum. The point of intersection, the degree of vulnerability, therefore, represents an outcome of the interaction of personal resources and environmental supports (Rogers, 1997, p. 68).

For young adults with pedLTC, increasing vulnerability occurs after transition. If their health condition was stable, changing symptoms may be noticeable. Comprehensive pediatric resources cease, and equivalent resources in the adult system are unavailable or difficult to attain. Subsequent lack of engagement with the health care system threatens their adherence to treatment and regimens, resulting in increased morbidity and costs in acute care settings during crises (Kennedy & Sawyer, 2008; Prestige, Romann, Djurdjev & Matsuda-Abedini, 2012). Further, long waits for specialist appointments for debilitating symptoms delays or precludes opportunities to achieve post secondary and vocational goals. Multiple, simultaneous and converging appraisals of the impact, threat, or challenge of changing system support, and their perception of the status of their condition, also influence their transition experience. The interplay of these factors impacts their navigation and negotiation of barriers and hurdles.

The Transition Engagement Rectangle encompasses several transitional factors: 1) the awareness and willingness to engage in the transition process, knowledge, beliefs and attitudes, and maturational level (Burke, Spoerri, Price, Cardosi & Flanagan, 2008; Lotstein, Ghandour, Cash, McGuire, Strickland & Newachek, 2009; Meleis et al., 2000); 2) resilience factors such as their

psychosocial, environmental, biological and cultural resources (Panter-Brick & Leckman, 2013); and 3) withdrawal from the process. The bi-directional arrows between the Vulnerability Triangle and the Transition Engagement Rectangle demonstrate that the degree of vulnerability experienced and resultant mastery, resilience, or withdrawal from the transition process are both interdependent and separate. For example, low vulnerability is likely associated with a higher level of mastery, but high vulnerability does not preclude the capacity for mastery. The individual's location within Transition Engagement Rectangle can slide laterally in either direction to accommodate and interact with the Vulnerability Triangle and Coping Appraisal Oval. Further, the continuum of mastery, resilience and withdrawal in the Transition Engagement Rectangle demonstrates one other important finding of this study. Two of the young adults who lacked environmental and personal resources such as community and family support, social connectedness, and opportunities to engage in meaningful activities appeared to use online gaming to numb or avoid these vulnerabilities. However, negative emotions cannot be selectively numbed; by default, both positive and negative emotions are numbed. Creativity, courage, and motivation to invest in uncertain possibilities were rooted in those who embraced their vulnerability.

The bottom of the drawing portrays the opposing and colliding forces of hoped-for developmental goals against a declining and plateauing health trajectory as time moves forward. While their health trajectory is beyond their control, the greater their degree of engagement in the new transition process and

their ability to engage resiliency factors, the more likely they will be to achieve their desired developmental outcomes. This diagram is not intended to suggest that these young adults are able to achieve whatever they desire, but rather, to demonstrate the requisite components that are most likely to support them in achieving their desired goals. Further, this diagram integrates two factors that were expected to be two incompatible: vulnerability and mastery.

Living with Uncertainty

Uncertainty pervades the experiences of young adults with pedLTC. Two substantial sources of uncertainty noted in this study were the impact of their condition, and the availability of resources and support from the adult health, education and social systems. The transition from pediatric to adult services heightened uncertainty and spawned a range of coping strategies and outcomes such as hopefulness and losing hope, realistic optimism, feeling depressed, and a pursuit of “finding normal.”

Uncertainty, in the context of illness, is multi-faceted and comprised of: ambiguity and lack of information about the condition, complexity of how to manage the condition within the care systems, and unpredictability about the trajectory and prognosis of the condition (Mishel & Braden, 1984). Hilton (1984) also attributed uncertainty to a lack of information, and added that the experience of uncertainty can be either threatening or positive. In this study, the young adults reported receiving adequate information in their pediatric years, but much less, especially in terms of anticipatory guidance, from their adult practitioners,

uncertainty and barriers navigating adult systems, and the unpredictability of their condition in the future. Relating the experiences of the young adults in this study to positive or threatening emotions, some framed their future expectations positively, even acknowledging that progressive decline and weakness were to be expected. Knowing that their condition was worsening, their positive outlook was supported by: 1) expectations that their condition would remain stable for the next 2-5 years, with minimal or moderate decline; 2) hopes for technological and medical advancements to cure or delay further progression of symptoms; 3) hope for technological support to compensate for their declining function; and 4) preferences to remain optimistic rather than contemplate uncertain and increasing unpleasant experiences.

For others, a lack of confirming information about their decline may serve as a protective mechanism, deflecting concerns about becoming progressively worse. However, for those who had undergone a tracheostomy to facilitate mechanical ventilation, the most important information to be shared with teens is to learn more about the procedure and what it is like to live with a tracheostomy before a medical crisis occurs. From their perspective, even though it is difficult to face when you are relatively stable, knowing your preference prior to young adulthood is imperative. Living in a zone of “not knowing” was not advised.

As a researcher, I experienced the participants’ preferences “not to go there” in thinking about difficult decisions. Recognizing their need for control and power over certain aspects of their lives, and that the research relationship is not

long term, I chose to be respectful of their reserve and not to probe for more information for the sake of knowledge on these difficult topics. In practice, it is important to ensure that space is provided for conversations about difficult topics in measured and safe amounts. Peer support in decision making for young adults is valuable.

In addition to inadequate information, other components of uncertainty in illness include ambiguity, inconsistency, vagueness, and unpredictability (McCormick, 2002). For pedLTC, ambiguity relates to discerning new or changing symptoms. Inconsistency may be associated with changing treatment modalities and recommendations for symptom management in the transition from pediatric to adult services, and the care providers' expertise and philosophy of care. Vagueness overlaps with ambiguity, focusing on the perception of the meaning of new and changing symptoms and information; Unpredictability is specifically related to the component of uncertainty related to prognosis, and not knowing what to expect in the future (Mishel, 1988). For those who were more reserved about their future, acceptance of one's declining function was predominant. Thus their appraisal of uncertainty as threatening or positive, and whether a lack of information related to ambiguity may determine the coping strategies they employ.

Finally, what uncertainty *is not* is the most interesting component related to pedLTC. Uncertainty is not about loss of personal control (Mast 1995). The young adults in this study and the video documentary participants (Talk About

Change, 2005) resonate with independence, despite their dependence on family or care aides. In this situation, like uncertainty, their appraisal of dependence as an opportunity and not a threat rings clearly.

End of Life for Young Adults with pedLTC

Embarking on this study, I anticipated lots of conversation with the participants about facing their end of life, but discovered that their conversations focused on present and future investments in their lives, finding meaning, searching for empowering strategies to achieve their goals, and having fun in the moment. And to be honest, end of life questions are hard to broach with young people. Many of the young adults in this study have “cheated death” several times, coming through tenuous medical circumstances on the side of life. So, is it unusual that they (like me) might be like Scarlett O’Hara, and prefer to think about it tomorrow? (Waples, 2012)

Preferences for end-of-life care were addressed in this study’s BBFG, although participant responses varied in their receptivity to discussion. A recent PhD study (Fletcher-Johnson, 2012) investigating future thinking of adolescents with Duchennes Muscular Dystrophy, also described wide-ranging levels of comfort discussing end-of-life topics. Her participants were portrayed as arriving at the end of adolescence with a “history of plateaus and crises” (p. 138) that predict a preference for “living in the moment”, and not preoccupied with potential “what ifs.” (p.138) Quoted previously in this current study, this participant’s comment substantiates “living in the moment”: “Truth is, I don’t

know, but either way, what is is, and I plan to keep a positive attitude.” In this study, questions about advance directives provided an estimation of their expectation that their health status may change unexpectedly. Nearly half of the participants did not know about advance directives, which may be as indicative of their current health status as their reserve to discuss them.

After setting up the BBFG, I came across four great end-of-life questions: Do you understand your prognosis? What are your fears about what is to come? What are your goals as time runs out? What trade-offs are you willing to make? Reflecting on this research experience, I hope to have further opportunities to discuss these questions with young adults with pedLTC - not to extract some sort of checklist for advanced directives, but to understand their hopes and fears, preferences, and support a peaceful end. And, beyond understanding their perspectives, I expect that their responses will teach and challenge me about my own hopes and fears, and my hard decisions, especially the unexpected ones (Waples, 2012). While shrouded by equipment and computers, living in wheelchairs and disfigured by the impact of their condition makes them hard to “be seen,” their wisdom and experience is illuminating.

Suggestions for Practice

Establishing New Initiatives in Practice Settings

Awareness of transition issues is growing as demonstrated by attention from professional bodies such as the Canadian (2013), American (2011), and United Kingdom (2008) pediatric societies. For example, in the United Kingdom,

an adolescent health e-learning training initiative for all health care providers has been developed (Royal College of Physician of Edinburgh, 2008), and the Canadian Association of Pediatric Hospitals (2012), has included transition as a new key initiative. However, the participant data confirms that it is the creative and time consuming work of the young adults, their families, volunteers, and some professionals that has initiated practice changes, need to be normalized into everyday practice across health, education and social systems (May et al., 2010).

With limited resources for the development of new programs and services, creating collaborative models of care through joint visits at transition to provide a comprehensive and multi-disciplinary hand-over from pediatric to adult specialists will acknowledge confidence in the adult providers and provide an opportunity for adult specialist training in the management of pedLTC.

Many authors have identified other specific initiatives to increase successful health transitions; these include certification requirements for professionals, and an up-to-date and portable health transfer summary and a comprehensive transition plan (Amaria, Stinson, Cullen-Dean, Sappleton & Kaufman, 2011; Shaw & DeLaet, 2010). These processes and initiatives will be advantageous for young adults and their families preparing for transition, and for their adult care providers. However, these individual factors are not sufficient to influence the outcome of complex system factors (extent of provider support, service availability) and personal factors (unique health, social and educational

needs, developmental readiness for self-management, and family support)
(Hamdani, Jetha & Norman, 2011).

Specialized Primary Care Practice

The complexity of managing progressively debilitating conditions, punctuated by stabilizing plateaus, and of understanding developmentally specific end-of-life needs and preferences, demands expert clinicians. Young adults with pedLTC require a safe passage from supportive pediatric and pediatric palliative care into adult services. While they are a heterogeneous group with diverse needs, they have much in common, and would benefit from an interdisciplinary team (Wein, Pery & Zer, 2010). However, as described previously, the expertise provided by pediatric teams in pedLTC, condition specific anticipatory guidance, and end-of-life support is absent in adult services. Further, in pediatric care, advance practice nurses manage and “hold” complex youth and their families together; in adult care there are few similar roles. The role of a primary care medical home is recommended in clinical guidelines and policy statements to provide continuity of care for complex and medically fragile patients (American Academy of Pediatrics, 2011; Brown, Katherine, Allen, Quach, Chiu & Bialystok, 2010). However, a recent report commissioned by the Canadian Health Services Research Foundation and the Canadian Nurse’s Association (Browne, Birch & Thabane, 2012, p. ix) determined the following components in clinical programs achieve the best outcomes for patients with chronic conditions and across the range of determinants of health:

1. Using nurse-led models of care (especially supplemental care models) that are proactive, comprehensive, coordinated and targeted, whether nurses are operating alone or as part of interdisciplinary teams that provide managerial continuity of care (This type of model entails a consistent and coherent approach from several professions to provide the agreed upon management of chronic, complex and changing patient needs.)
2. Integrating nurse-led models of care with interdisciplinary teams that are based on an ecological understanding of the interplay among a myriad of personal and environmental factors determining patients' health (These complex interventions require a high-quality primary healthcare system and patient-centered care practices led by specially trained nurses or advanced practice nurses as well as adequate investments in social programs.)
3. Working within a system where the amount of money spent on social services is higher than that spent on health services (The ratio of social service expenditures to health service expenditures is associated with better outcomes in key health indicators in countries belonging to the Organization for Economic Co-operation and development.)

Development of specialty and multi-disciplinary practices and resources in primary care is requisite to provide supportive care and knowledge for young adults with pedLTC is. Further, to understand the meaning and implications that

ongoing losses have for young adults with pedLTC, skilled and expert clinicians are required. As demonstrated in this study, threats, harm and loss can be fuel for engagement and change, or result in retreat and subsequent loss of social connectivity and meaningful engagement.

Young Adult Hospice

In addition to specialized and multidisciplinary primary care practices, young adult hospice (YAH) - which is gaining momentum in the United Kingdom to support this new and growing population of young adults with pedLTC - is also a worthy consideration. Currently, hospice and palliative services focused on pedLTC and young adult developmental needs are not available outside of the United Kingdom. YAH homes and hospice societies can support successful transition from pediatric to adult services, independence, social networks, recreation, end of life decisions and care; and provide specialized symptom control, psychosocial and spiritual resources, and access to work and educational opportunities. Expert knowledge about symptom management and end of life trajectory, a trusting relationship with adult care providers skilled in end of life planning, and a coordinated plan for medical emergencies, are essential components of support services for young adults with pedLTC. Gaining the trust and respect of young adults, who have had ample opportunities to observe insincerity and lack of clarity in their health providers, is an essential attribute of hospice clinicians. Ideally, dying in a hospice would be with dignity and in the “presence of people who know how to drop the professional role mask and relate

to others simply and richly as a human being” (Roy, 1986 in Wein, Pery & Zer, 2010, p. 4819).

However, there are intricacies and complexities in YAH care that require careful consideration. By virtue of the diverse population requiring YAH services, creating a common philosophy and mandate for YAH is complicated. For example, young adults with life limiting conditions are comprised of at least three distinct groups: 1) the cognitively capable young adults with pedLTC described in this study, 2) non-cognitively competent young adult with pedLTC, and 3) those with life threatening conditions that are diagnosed in young adult years (e.g. cancer). Between the cognitively and non-cognitively competent groups, the types of activities enjoyed, and level of attendant care will vary widely. In a physical hospice structure, where young adults stay for respite, symptom management, or end-of-life care, differences in cognitive competency will create complications providing activities and events that are of interest, or appropriate to everyone. Further, prognostication of the trajectory of life threatening conditions is not an exact science. In pediatric hospice care, the mandate for care provision ends at 18 or 19 years of age. In YAH, the years of care provision are unknown as young adults live longer with pedLTC.

Contributions to Nursing Practice

Pediatric nurses in tertiary centers frequently are “the glue” for families and youth as they navigate and negotiate for needed services in their pediatric years; and sometimes they continue to be a constant support into their adult years.

The participants in this study did not mention similar relationships with adult providers; likely because there are fewer multidisciplinary teams in adult practice. It is worth considering the components of health provider behaviors that were described as the most helpful by the young adults for both pediatric and adult providers.

Forming a collaborative relationship with patients is the essence of the nursing relationship, and pivotal for client empowerment and the successful achievement of their desired outcomes (Gallant, Beaulieu & Carnevale, 2002; Gottlieb & Feeley, 2005). As described by these young adults, feeling understood and known by their care providers are attributes that are highly valued (Allison, Stacey, Dadds, Roeger, Wood & Martin, 2003; Doane & Varcoe, 2005; Gottlieb & Feeley, 2005). However, the biomedical and service provision models of health care have historically shaped a different kind of nurse-patient relationship. The biomedical model is based on the premise that professional expertise superseding client perspectives; an approach that allocates more power to the provider, than the client (Alison et al.; Brown, McWilliam & Ward-Griffin, 2006; Doane & Varcoe). And the service provision model focuses on deficiencies, health problems and how to “fix” them (Doane & Varcoe). In pediatric care (both acute and palliative), health care providers have a tendency to “do for” children and youth, rather than using their expertise and knowledge to assist the youth to explore and evolve into their role as partners in care (Brown et al.). In adult care, interdisciplinary teams with nurses are less common, resulting in fragmented and

uncoordinated care for young adults with pedLTC. Reflection on the models of care influencing nursing practice, and research to inform partnering and empowering approaches to care is needed.

Another important finding in this study was the young adults' surprising capacity for resilience. The biomedical problem-oriented approach focuses on problems, and results in feeling helpless, despondent, and unable to identify hoped for goals and achievements (McAllister, 2003). Conversely, nursing practice that is rooted in a strengths based orientation, focuses on strengths and capabilities that have enabled successful navigation of previous difficulties, and support to search for alternate solutions to unsuccessful strategies (Black & Lobo 2008; Alison et al, 2003; McAllister). A solution-focused orientation problem solves with the clients, not for them, and helps clients build on their achievements (McAllister).

Finally, community-based and nurse led interdisciplinary teams that include a primary care physician are key components of effective and efficient provision of complex care to a complex group of patients. Multi-faceted interventions that include both health and social interventions require specially trained or advanced practice nurses to supplement primary care physicians and other health care professionals. This coordinated model of care provides proactive and comprehensive community care to vulnerable populations with complex health conditions and social circumstances (Browne et al., 2012).

Strengths and Limitations of this Study

First and most importantly, this study is an exploration of two uncharted territories: the experiences of young adults with pedLTC living into adulthood, and the development, implementation and evaluation of a novel research strategy to access medically fragile populations. This study has provided a foundational description of the barriers, challenges, and opportunities in the health, education and social service systems from the perspective of the young adult, and describes one of the first attempts to employ an innovative research method to an otherwise inaccessible population.

Original Contributions to Research

First, this study provides an original contribution to the theory of coping with and appraisal of stress (Lazarus & Folkman, 1984). Many stressors related to the complexities of pedLTC and transition superimpose on each other, and multiply in effect for these young adults. It was demonstrated that not only is the stressor appraised as a threat, harm or loss, it can be viewed as 1) each of these distinctly, 2) all of them together, and/or 3) all of these as a challenge to overcome. To date, stress appraisal has been viewed as single measure, rather than multiple concurrent and converging appraisals.

Second, the multiple and converging realities of these young adults including uncertainty, vulnerability, resilience, and loss and grief superimposed on a young adult developmental trajectory supports a meta theory of paradox. Their reality co-exists and is in constant re-appraisal as each event is measured

and weighed against their personal resources, environmental supports and hoped for dreams and passions.

Third, the method of data collection gave voice to those previously unheard and is a major contribution to nursing intervention strategies and opportunities for meaningful young adult engagement. Finally, this thesis demonstrates the value of narrative and cognitive behaviour strategies not usually considered as transition interventions.

Methodological Strengths

Triangulation. In this case study, methodological rigor was preserved and enhanced through iterative triangulation of the data at all junctures of the study from the selection of data sources to the review of written manuscripts. Multiple data sources, including young adults with pedLTC, relevant clinical experts, and documents; and two different types of interview methods enhanced the quality of the data and facilitated a rich understanding of their experiences (Creswell, 2007; Yin, 2009). Case study methodology also facilitates triangulation through a synchronous process of data collection and analysis, allowing comparison of data with theory, and opportunities for further investigation and understanding of the findings in every step of the research process (Yin). Further, content analysis provides a framework to support triangulation of the multiple data sources used in this study (Hsieh & Shannon, 2005).

Credibility. The most important criterion by which to judge qualitative research is its credibility. Credibility and confidence in the findings were ensured

through prolonged engagement with the participants and data in both the face-to-face and BFG interviews, persistent observations, memo writing, member checking and searching for disconfirming evidence (Creswell, 2007; Lincoln & Guba, 1985). Participants and clinical experts provided descriptive and contextual verification of the study results from their perspectives. For example, one participant commented:

I particularly liked your descriptions of the transition from child to adult as an "abyss" and "like falling off a cliff".... I think those are strong & accurate metaphors for what we described in group. You mention vulnerabilities and resiliency in the very beginning and I really liked how the words (vulnerability and resiliency) were kind of weaved through out the entire paper as it does encapsulate the theme and tie it all together.

And a clinical expert from CPCH commented: “This is comprehensive and has great insights. Examples were powerful and the issues well covered . . . So affirming of what I witness clinically ” (S. Poitras, personal communication, June 13, 2013)

Dependability and confirmability. Double coding of initial interviews with two supervisory committee members; and iterative refining of the codes and meaning of the data as more information was collected from subsequent participants, and compared with the opinions of clinical experts, enhanced data dependability and confirmability. Other dependability measures in this study included: 1) a detailed research process that was critiqued and vetted by academic

supervisors and two university ethics boards; 2) reviewing the memos written after each face-to-face interview and, pre, mid and post BBFG groups with two academic advisors also expanded my perspectives and hunches about the unfolding data and; 3) detailed audit trails of the development of the face-to-face and BBFG interviews (Miles & Huberman, 1994).

A potential risk to confirmability in this study is the multiple roles undertaken the researcher: interviewer, BBFG moderator and observer, and data analyst. This risk was mitigated through training in family therapy that restrains clinical and research assumptions to focus on following the participants' stories. Conversations with the BBFG group about the meaning of their online activity, and being dependent enabling their independence, are examples where following the participants' leads overrode my assumptions.

Transferability. Transferability of the findings is supported by the rich descriptions of the participants and the research context, and confirmation of the findings by the participants, and clinical experts who observe and work closely with a broad range of young adults with pedLTC.

Methodological Limitations

Limitations of this study include a relatively small sample size of young adults with pedLTC, though they were chosen to be representative of others with similar conditions. Transferability of the data is limited to cognitively capable young adults with pedLTC. Cognitively competent young adults with pedLTC comprise roughly half of all young adults with pedLTC. While the intent of this

study was the young adult's perspective of their transition experience, their interconnectivity with their family, and health, education and social systems requires the perspective of these stakeholders. Parents or care providers in extended care homes and hospitals provide increasing care and support as the health status of these young adults declines.

Innovative Research Methods Strengths and Limitations

The proliferation of social media, online discussions and text conversations make online modalities familiar and comfortable for young adults. The greatest strengths of the BBFG are the appeal of this methodology for young adults, and the multi day focus group becomes both a community and an intervention. The diversity of physical limitations and communication modalities among the participants in this study demonstrates the inclusiveness and adaptability of an asynchronous BBFG forum for medically fragile populations. The participants in this study, like those cited in a recent review of the literature (Sansom-Daly, Peate, Wakefield, Bryant & Cohen, 2012), are highly amenable to participating in an online discussion. While there were instances of limited follow-through by some participants on the BBFG discussion threads, the resulting data were rich and creative. Finally, engaging young adults with pedLTC in the development and support of online focus group provides meaningful engagement, opportunities to “give back,” and mentor those who are younger.

Further Research

Systems

Against the dearth of research regarding the experiences of young adults with pedLTC who have transitioned from pediatric to adult care, this research focused on systems issues from the young adults' perspectives. Further research that encompasses all the stakeholders - young adults, parents, and health, education and social service providers - will clarify the needs and priorities of each group. A PhD thesis currently in progress in Northern Ireland is addressing all these stakeholders to determine the opportunities and challenges for providing better care to young adults with pedLTC within their geographical limits (H. Kerr, personal communication, May 22, 2013). In Canada, investigation of current provincial strategies and resources in the health, education and social sectors will determine the overlap and gaps in the allocation of services. Further understanding of health system priorities, current specializations and services in pediatric and adult palliative and hospice care, and opportunities for collaboration between pediatric and adult services will enhance understanding of the web of support and services required for young adults with pedLTC.

Young Adults

Further research is needed to 1) understand the interconnectedness of personal characteristics, appraisal and environmental supports for young adults with pedLTC, and 2) discern how to empower these resources and modulate stress appraisal to enhance resilience and developmental life achievement. Research that explores relationships among coping strategies and resiliency, hope, social

support and personality characteristics will help to illuminate the dual appraisal of threat and hopefulness in those living with pedLTC. More research is also required to understand how to support intimacy, and to determine and honour preferences for end of life decisions and care.

Online Research

Continued testing and refinement of online methods with other groups of hard-to-reach participants, more feedback about participants' experiences will continue to add merit to BBFG methodology. Given the ease of participating in online groups, research that considers the dual benefits of online groups as research and intervention is warranted. Further research is needed to determine the efficacy of online support groups, and whether peer or professional moderators or a combination is the best. The quality of data generated remains dependent on the skills of the researcher and moderator in building an engaging, personalized and coherent discussion over several days using varied mediums. Finally, engaging young adults with pedLTC in the development and support of an online focus group provides meaningful engagement, as well as opportunities to “give back” and mentor those who are younger.

Final Thoughts

Call to Action

Mike is 30 years old with Duchenne's Muscular Dystrophy, has chronic pain, “lives” in his power wheelchair, is ventilated, requires feeding support, and his condition is declining. He has been living in an intermediate care home for the

past 5 years. As his symptoms change and escalate, he is being bounced between adult specialist consultants who provide a “patchwork” of care. While his family doctor is attentive, he does not have the support of an allied health team to coordinate Mike’s care, or the expertise to provide expert medical management of his symptoms and needed anticipatory guidance. The intermediate care home limits the number of requests for help allowed each night, and he now exceeds their quota. He is in pain, and in crisis because he has to leave his home at a time when what he needs most are the comfort and security of familiar surroundings and support. Mike is “unseen” in the system, yet shoulders all of the responsibility for managing his care. He will not be eligible for adult hospice services because his end of life trajectory cannot be prognosticated within the required time frame. He will likely have a precipitous end in an acute care or intensive care hospital setting, disconnected from his friends, family and things he loves most (S. Poitras, personal communication, June 13, 2013).

Mike is real, and his current situation demonstrates how the systems fail young adults with pedLTC, and also provides a future view for participants in this study who may be unable to receive enough support to live independently or at home with their aging parents. As their condition declines, young adults with pedLTC will experience diminishing energy and function, thus lessening their capacity to draw on their resilience and coping strategies. Provider, program, system and societal changes are required to change the fate of these unprotected and vulnerable young adults.

In Conclusion

Qualitative research has been called “data with a soul.” (Brown, 2010)

This study reveals a depth of humanity seldom seen and experienced by few. Experiencing the stories of these young adults moved me far beyond the data and the research process; it was profound and I am wholly grateful. I am indebted to all of my participants for opening up their lives, and sharing their stories, hopes, disappointments, and vulnerability – the source of their courage and creativity - with their fellow participants and me. My hope is that through honoring and telling what is “ordinary” in your lives, providers, programs, and systems will be transformed to support your extraordinary journeys.

For me, Victor Frankl (1946) sums up the essence of this study, and the reason to work for changes needed to support this vulnerable group of outstanding young women and men.

Life is never made unbearable by circumstances, but only by lack of meaning and purpose.

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Appendix A: Combined Ovid, CINAHL and Cited Reference Search Results



Restrictions: 2000 – 2010, English, Human

Search Terms: Transition*; Transition* to adult health care; care; Adolescent transition*; Transition from pediatric to adult care

Results:

	Number of Articles
Diseases of the endocrine, nutritional, metabolic systems	1
Nervous system diseases	3
Circulatory system diseases (Congenital heart defects)	11
Respiratory system diseases (Cystic fibrosis)	13
Digestive system	0
Others: Transplant, sickle cell, spina bifida, genetic anomalies, diabetes, cerebral palsy, hydrocephalus, HIV/AIDS, transplant	14
Consensus and policy statements; general standards; models of care	18
Health practitioner and family perspectives	5

Appendix B: Face-to-Face Interview Recruitment Letter and Consent Form



LETTER OF INFORMATION / CONSENT (Interviews)

A study about the transition experience of young adults from Canuck Place Children's Hospice (Vancouver, BC) to adult services

Investigators:

Principal Investigator:
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Funding Source: McMaster Faculty of Health Sciences, School of Nursing, and CIHR Advanced Practice Nurse Research Chair


Purpose of the Study

The purpose of this study is to explore the experiences of young adults who have transitioned from pediatric palliative care services at Canuck Place to adult services. This research is being conducted because there are no programs that help youth transition from pediatric palliative care to adult services, and the care provided to young adults with pediatric life limiting conditions needs to be improved. One way to improve the care is to find out from young adults like yourself what is going well and what needs improvements and changes. Your assistance with this study will help to improve the services and care for other young adults like yourself in the future. This research study is part of Karen Cook's doctoral studies.

What will happen during the study?

Two young adults who have had at least 3 visits to Canuck Place during their teen years will participate in 2 to 3 face-to-face interviews, each lasting about an

Version August 15, 2011



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hour. The interviews will be held at mutually convenient times and location and will be conducted by the study's principal investigator, Karen Cook. Prior to starting the interviews, you will be asked for some background information like your age and education, who is in your family, and your medical history. During the interviews, you will be asked questions about your experiences living with, and managing your complex pediatric condition since you left Canuck Place and entered adult services. With your permission, these interviews will be digitally recorded and transcribed by another member of the research team.

To fully appreciate your medical history and your care at Canuck Place Children's Hospice, the researcher will ask permission to review your Canuck Place Children's Hospice health record.

Are there any risks to doing study?

It is not likely that there will be any harms or discomforts associated with answering the interview questions. This research topic deals with your own experiences living with and managing your complex pediatric condition during the time you left Canuck Place and entered adult services. You do not need to answer questions that you do not want to answer or that make you feel uncomfortable. You may decline further participation at any time. If you feel uncomfortable in any way from answering the interview questions, you will be provided with the name and contact information of a counselor who will be happy to speak with you. On the other hand, some people find it beneficial to talk to others about their own experiences and learn from other's experiences.

Are there any benefits to doing this study?

What is learned as a result of this study will help to provide better services to young adults like yourself who transition from pediatric palliative care services, like Canuck Place, to adult services.

Payment or Reimbursement

In appreciation for the time you give to the interviews, you will receive a \$25.00 gift card to iTunes or Starbucks. In the event there are travel or parking expenses, they will be reimbursed.

Confidentiality

Every effort will be made to protect (guarantee) your confidentiality and privacy. Your name and any information that would allow you to be identified will not be used. However, we are often identifiable through the stories we tell. The digital



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recordings will be erased when the research is completed. All identifying information will be removed from the data. Code numbers will be used to identify all personal information in documents, and these documents will be kept in a locked cabinet where only the researcher will have access. Information kept on a computer will be protected by a password. Identifying information will not be emailed to anyone at any time. You will not be identified by name in any reports of the completed study. Only study personnel will have access to the study data. All of the files with identifying information will be destroyed 10 years after the study is complete.

What if I change my mind about being in the study?

Your participation in this study is voluntary. It is your choice to be part of the study or not. If you decide to be part of the study, you can decide to stop (withdraw), at any time, even after signing the consent form or part-way through the study. If you decide to withdraw, there will be no consequences to you. In cases of withdrawal, any data you have provided will be destroyed unless you indicate otherwise. If you do not want to answer some of the questions you do not have to, but you can still be in the study. Your decision whether or not to be part of the study will not affect your continuing access to any services you currently receive.

How do I find out what was learned in this study?

It is expected that this study will be completed by approximately September 2012. If you would like a brief summary of the results, please identify how you would like it sent to you.

I will receive a signed copy of this form.

Questions about the Study

If you have questions or need more information about the study itself, please contact Karen Cook:
karencook8@gmail.com
604 345 8915

This study has been reviewed by the Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board and the University of British Columbia's Research Ethics Board and received ethics clearance.

If you have concerns or questions about your rights as a participant or about the way the study is conducted, please contact:

The Office of the Chair of the Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board
Telephone: (905) 525-2100 ext. 42013
E-mail: ethicsoffice@mcmaster.ca



CONSENT

I have read the information presented in the information letter about a study being conducted by Karen Cook of McMaster University.
I have had the opportunity to ask questions about my involvement in this study and to receive additional details I requested.
I understand that if I agree to participate in this study, I may withdraw from the study at any time. I have been given a copy of this form. I agree to participate in the study.

Signature: _____

Name of Participant (Printed) _____

Date: _____

1. I agree that the interview can be audio/video recorded.
... Yes.
... No.

2. I agree that the researcher may review my Canuck Place Children's Hospice health record.
... Yes
...No



3. ...Yes, I would like to receive a summary of the study's results.
Please send them to this email address _____
or to this mailing address: _____

... No, I do not want to receive a summary of the study's results.

4. I agree to be contacted about a follow-up interview, and understand that I can always decline the request.
... Yes. Please contact me at: _____
... No.



Appendix C: BBFG Recruitment Letter and Consent Form

SUBJECT INFORMED CONSENT FORM (Online Focus Groups)

A study about the transition experience of young adults from Canuck Place Children's Hospice (Vancouver, BC) to adult services

Investigators:

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Funding Source: McMaster Faculty of Health Sciences, School of Nursing, and
CIHR Advanced Practice Nurse Research Chair

Purpose of the Study

Version 2, July 26, 2012



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You are invited to participate in a study to explore the experiences of young adults who have transitioned from pediatric palliative care services at Canuck Place to adult services. This research is being conducted because there are no programs that help youth transition from pediatric palliative care to adult services, and the transition services provided to young adults with pediatric life limiting conditions needs to be improved. One way to improve the care is to find out from young adults like yourself what is going well and what needs improvements and changes. Your assistance with this study may help to improve the services and care for other young adults like yourself in the future. This research study is part of Karen Cook's doctoral studies.

What will happen during the study?

Six to ten young adults who have had at least 3 visits to Canuck Place during their teen years will participate in an online focus group that is moderated by the study's principal investigator, Karen Cook. The online focus group is accessible from your computer and you will be provided with the software and operating support that you require to participate in the online focus group. You will be asked to log onto the private online focus group at least twice a day for 5 days. In the event that you are unable to participate during this time, the online focus group can remain open to allow you to participate. You will be asked questions about your experiences living with, and managing your complex pediatric condition since you left Canuck Place and entered adult services. You will be able to post your own responses to the questions, and respond to the comments posted by the other participants.

As part of the online focus group, you will also be asked for some background information like your age, what you are doing now, who is in your family, and accessing adult services. This information can be collected online or via telephone, depending on your preference.

To fully appreciate your medical history and your care at Canuck Place Children's Hospice, the researcher may ask permission to review your Canuck Place Children's Hospice health record.

Are there any risks to being in the study?

It is not likely that there will be any harms or discomforts associated with answering the interview questions. This research topic deals with your own experiences living with and managing your complex pediatric condition during the time you left Canuck Place and entered adult services. You do not need to answer questions that you do not want to answer or that make you feel



uncomfortable. You may decline further participation at any time. In case you feel uncomfortable in any way from answering the questions in the online focus group, the name and contact information of a counselor who will be happy to speak with you, will be provided to you before the study starts. On the other hand, some people find it beneficial to talk to others about their own experiences and learn from other's experiences.

Are there any benefits to doing this study?

What is learned as a result of this study may help to provide better services to young adults like yourself who transition from pediatric palliative care services, like Canuck Place, to adult services.

Payment or Reimbursement

In appreciation for the time you give to the interviews, you will receive a \$25.00 gift card to iTunes or Starbucks. In the event there are travel or parking expenses, they will be reimbursed.

Confidentiality

While every effort will be made to protect your confidentiality and privacy, the nature of focus groups prevents a guarantee of confidentiality. All participants will be reminded to respect the privacy of their fellow participants and not repeat what they read on the online focus group. You may choose to identify yourself using a name that is your own or not while you are online.

Your name and any information that would allow you to be identified will not be used. However, we are often identifiable through the stories we tell. All identifying information will be removed from the data. Code numbers will be used to identify all personal information in documents, and these documents will be kept in a locked cabinet, which is only accessible to the researcher. Information kept on a computer will be protected by a password. Identifying information will not be emailed to anyone at any time. You will not be identified by name in any reports of the completed study. Only study personnel will have access to the study data. All of the files with identifying information will be destroyed 10 years after the study is complete.

The private and secure online focus group will be set up using "iTracks". This is a Canadian company that services more than 2500 online groups every year. All



data is stored and accessible only in Canada. The online focus group is only accessible with a unique code and password that is provided by iTracks. The only people able to read the questions and comments posted on the focus group are the investigator, a supervisory committee member, and the participants. Data collected through the online focus group is protected through the comprehensive security measures of the iTracks software system which includes the hardware, software, and data security and storage. Detailed information about the security measures of iTracks will be provided at your request.

What if I change my mind about being in the study?

Your participation in this study is voluntary. It is your choice to be part of the study or not. If you decide to be part of the study, you can decide to stop (withdraw), at any time, even after signing the consent form or partway through the study. If you decide to withdraw, there will be no consequences to you. If you do not want to answer some of the questions you do not have to, but you can still be in the study. Your decision whether or not to be part of the study will not affect your continuing access to any services you currently receive. To withdraw, you can stop participating in the online focus group and email the researcher, Karen Cook, to notify that you have withdrawn.

How do I find out what was learned in this study?

It is expected that this study will be completed by approximately September, 2012. If you would like a brief summary of the results, please identify how you would like it sent to you.

You will receive a signed and dated copy of this form.

Questions about the Study

If you have questions or need more information about the study itself, please contact Karen Cook:
karencook8@gmail.com
604 345 8915

The Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board and the UBC/Children's and Women's Health Center of BC Research Ethics Board have reviewed and provided ethics clearance for this study.



If you have any concerns or complaints about your rights as a research subject and/or your experiences while participating in this study, contact the Research Subject Information Line in the University of British Columbia Office of Research Services by email or at RSIL@orsubc.ca or by phone at 604 822 8598 (Toll free: 1-877-8222-8598).

AND/OR

The Office of the Chair of the Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board, Telephone: (905) 525-2100 ext. 42013, E-mail: ethicsoffice@mcmaster.ca



CONSENT

I have read the information presented in the information letter about a study being conducted by Karen Cook of McMaster University. I have had the opportunity to ask questions about my involvement in this study and to receive additional details I requested. I understand that if I agree to participate in this study, I may withdraw from the study at any time. I have been given a copy of this form. I agree to participate in the study.

Signature: _____

Name of Participant (Printed) _____

Date: _____

Name of assistant (if required) for online focus group:

Signature: _____

1. I would like to receive a summary of the study's results.

... Yes, Please send them to this email address

Or to this mailing address:

... No, I do not want to receive a summary of the study's results.



2. I agree that the researcher may review my Canuck Place Children's Hospice health record.

... Yes
... No



3. I agree to be contacted for follow-up questions, and understand that I can always decline the request.

... Yes, Please contact me at: _____

... No



Appendix D: Specialist Informant Recruitment Letter and Consent Letter

HEALTH CARE AND COMMUNITY PROVIDER INFORMED CONSENT FORM

A study about the transition experience of young adults from Canuck Place Children's Hospice (Vancouver, BC) to adult services


Investigators:

Principal Investigator:
Dr. Hal Siden
Clinical Associate Professor
Department of Medicine, UBC

Co-Investigator
Karen Cook, BSN, MSc, PhD (student)
Visiting Professor, UBC School of Nursing,
And
Assistant Professor, School of Nursing,
McMaster University
Hamilton, Ontario, Canada
604 345 8915
E-mail: karencook8@gmail.com

Faculty Supervisor: Dr. Gina Browne, Ph.D., Reg.N., Hon.LL.D
Founder and Director, Health & Social Service Utilization Research Unit
Professor, Nursing; Clinical Epidemiology & Biostatistics; and
Ontario Training Centre in Health Services and Policy Research (OTC)
MOHLTC Theme Lead - Innovative and Integrated Systems of Prevention and
Care
McMaster University - McMaster Innovation Park
175 Longwood Road South, Suite 210A, Hamilton, ON, Canada L8P 0A1
Telephone: 905-525-9140, Ext. 22293 Fax: 905-528-5099
E-mail: gina.browne@mcmaster.ca
Website: www.fhs.mcmaster.ca/slru/home.htm

Funding Source: McMaster Faculty of Health Sciences, School of Nursing, and
CIHR Advanced Practice Nurse Research Chair

Version January 30, 2012  Page 1 of 6



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Purpose of the Study

You are invited to participate in a study to explore the experiences of young adults who have transitioned from pediatric palliative care services at Canuck Place to adult services. This research is being conducted because there are no programs that help youth transition from pediatric palliative care to adult services, and the care provided to young adults with pediatric life limiting conditions needs to be improved. One way to improve the care is to find out from the young adults and their care providers what is going well, and what needs improvements and changes. This research study is part of Karen Cook's doctoral studies.

What will happen during the study?

An interview, lasting approximately one hour will be scheduled at a mutually convenient time and location, and will be conducted by the study's principal investigator, Karen Cook. Prior to starting the interviews, you will be asked for some background information like your age, specific job title and profession, number of years you have worked with this population, and the setting of your practice (primary care, community, hospital etc.) During the interview, you will be asked questions about your experiences working with youth with pediatric life limiting conditions who have transitioned to adult care. With your permission, these interviews will be digitally recorded and transcribed by another member of the research team.

Are there any risks to being in the study?

It is not likely that there will be any harms or discomforts associated with answering the interview questions. This research topic deals with your own professional experiences. You do not need to answer questions that you do not want to answer or that make you feel uncomfortable. You may decline further participation at any time.

Are there any benefits to doing this study?

What is learned as a result of this study may help to provide better services to young adults who transition from pediatric palliative care services, like Canuck Place, to adult services.

Confidentiality

Every effort will be made to protect (guarantee) your confidentiality and privacy. Your name and any information that would allow you to be identified will not be used. However, we are often identifiable through the stories we tell. The digital



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recordings of the interviews will be transcribed by a professional transcriptionist and will be erased when the research is completed. Participants will be provided with code names and all identifiers will be stripped from the transcripts and field notes. Code numbers will be used to identify all personal information in documents, and these documents will be kept in a locked cabinet where only the researcher will have access. Information kept on a computer will be protected by a password. Identifying information will not be emailed to anyone at any time. You will not be identified by name in any reports of the completed study. Only study personnel will have access to the study data. All of the files with identifying information will be destroyed 10 years after the study is complete.

What if I change my mind about being in the study?

Your participation in this study is voluntary. It is your choice to be part of the study or not. If you decide to be part of the study, you can decide to stop (withdraw), at any time, even after signing the consent form or part-way through the study. If you decide to withdraw, there will be no consequences to you. In cases of withdrawal, any data you have provided will be destroyed unless you indicate otherwise. If you do not want to answer some of the questions you do not have to, but you can still be in the study.

How do I find out what was learned in this study?

It is expected that this study will be completed by approximately September 2012. If you would like a brief summary of the results, please identify how you would like it sent to you.

You will receive a signed and dated copy of this form.

Questions about the Study

If you have questions or need more information about the study itself, please contact Karen Cook:
karencook8@gmail.com
604 345 8915

The Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board and the UBC/Children's and Women's Health Center of BC Research Ethics Board have reviewed and provided ethics clearance for this study.

If you have any concerns or complaints about your rights as a research subject and/or your experiences while participating in this study, contact the Research Subject Information Line in the University of British Columbia Office of Research



1-877-822-8598



CHILD
& FAMILY
RESEARCH
INSTITUTE

Services by email or at RSIL@orsubc.ca or by phone at 604 822 8598 (Toll free: 1-877-8222-8598).

AND/OR

The Office of the Chair of the Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board, Telephone: (905) 525-2100 ext. 42013, E-mail: ethicsoffice@mcmaster.ca



CONSENT

I have read the information presented in the information letter about a study being conducted by Karen Cook of McMaster University.
I have had the opportunity to ask questions about my involvement in this study and to receive additional details I requested.
I understand that if I agree to participate in this study, I may withdraw from the study at any time. I have been given a copy of this form. I agree to participate in the study.

Signature: _____

Name of Participant (Printed) _____

Date: _____

1. I agree that the interview can be audio/video recorded.

- ... Yes.
- ... No.

2. I would like to receive a summary of the study's results:

... Yes

Please send them to this email address:

OR to this mailing address:

... No, I do not want to receive a summary of the study's results.



Faculty of Health Sciences



CHILD
& FAMILY
RESEARCH
INSTITUTE

3. I agree to be contacted about a follow-up interview, and understand that I can always decline the request.

... Yes. Please contact me at: _____

... No.



Appendix E: Interview Guide for Young Adult Face-to-Face Interviews

The following questions serve as a topical guide and prompt for interviews with youth with pediatric life limiting conditions and are not intended as an exhaustive list or a schedule to be rigidly followed. Conversation and questions will follow leads provided by the participants in response to the types of questions listed below. These questions and conversations will occur over two to three sessions.

All About Yourself

1. Tell me about yourself.
 - Who is in your family?
 - Where do they live? What are they doing?
 - How long have you lived by yourself/with family/support worker?
 - What are you doing now?
 - School, work, volunteer, other things

2. What do you like to do? Tell me more about . . .
 - Can you imagine being able to do _____?
 - What would it take for you to be able to do _____?
 - Who/what do you think would be able to help you to do _____?

Impact of Health Condition on Transition

3. Tell me about your medical condition
 - How long have you lived with (illness)?
 - What have you learned about yourself living with _____?
 - What gives you the strength to live with _____? Does anything or anyone help you have the strength when you don't feel like it?
 - What parts of your medical condition can you take care of yourself?
 - Who helps you with what you can't do? Are they able to help you in a way that works for you? What could be different/better?
 - Has your (medical condition) changed since you were a child/teen ager? How?
 - What do you think your (medical condition) will be like in 2 years? 5 years? Longer?
 - Who/what would you like to have/need to manage (medical condition) in next 2 – 5 years?

- Do you know how you will get these things/people? Do you know anyone who can help you?
- What do you think is important for teens to know about and prepare for when they have _____? If you had the opportunity, what would you want to talk to them about?

Transition

4. One of the big changes that I know about that happens when you make the transition from child/teen services to adult services is that some resources are harder to get. By resources I mean, specialists that understand your condition (like nurses, doctors, counsellors, physios), money (for medical supplies, living, school, transportation), support to go to school, and making connections with your friends and other youth that you know and understand you. Tell me about:

Health

- Who are your health care providers? (people who help with your medical condition?)
- How did you find these health care providers?
 - Do you feel that they understand you and your condition?
 - Do any of these health care providers help you get services or care that you or your family can't get on your own?
- Describe what it is like getting to your appointments.
 - On a scale of 1 – 10, rank your motivation to go, difficulty/ease of seeing everyone you are referred to see.
 - Do any of these health care providers come to see you?
 - Do any of these health care providers have offices in the same clinics or hospital so that you don't have to get to different places?
 - Is this different/same/worse/better than when you went to medical appointments in children/teen services? Tell me more.
 - If you could design a program that helped other people with a similar condition to yours, how would it work?

Education

- Have you been able to finish high school?
- What was it like for you in high school when your classmates were preparing for their futures: school, work, travel, and you were aware that your future would likely be much shorter? What did you want to do?
- Are you or have you taken any courses or programs at a college or university?
 - How are you able to do this?
 - What assistance (financial/accessibility/accommodations) do you need?
 - Who has helped you or What help will you need to be able to do this? Will you require further help to carry on? Do you know how you will get this help?

Independent Living, Transportation, Work, Social Connections

- Similar questions to above
5. Another big change that happens when you make the transition from child/teen services to adult services is learning how to advocate, or get what you need for yourself. Some other youth have told me that in their child and teen years, their parents or health professionals (or others) decided what was best. As they became young adults, they realize that while their parents/health professionals thought they knew what was best, the youth had to figure out what they really wanted for themselves and how to get it.
- Has this been similar for you? Tell me more about this . . .
 - Has there been a time when your wishes have been quite different from your parents or health care providers? Tell me more . . .
 - You are reliant on people to help you manage your medical condition. How are you able to be both reliant on people for your care, and stand up for what you want?
 - What has helped you to be able to do this?
AND/OR What makes it hard to do this?
6. What other changes happen when you move from child/teen services to adult services. Tell me more about _____.
7. Tell me what you remember about getting ready to “move or transition” from Canuck Place? / Children’s Hospital / services that are for children and teens only?

- What did you learn about? How did you learn these things?
- How did you learn about how to get the health care services you needed? Money for transportation and living? Money for/access to equipment? Possibilities for taking courses/work/volunteer?
- Do you remember how old you were when you started learning about all that is involved in transitioning from pediatric to adult care? Did you want to think/learn about this at the time? Tell me more about this.
- Was any of this helpful to you? What was most/least helpful? Tell me more.
- What could be different? What suggestions do you have for the people that help teens get ready to transition from children and teen services to adult services?
- If you were asked to talk to a teenager who is similar to yourself, what would you talk to them about the transition from children/teen services to adult services?
- Is there anything you would do differently in your teen years knowing what you know now?

Young Adult Years

8. What are the most important things in your life right now?
 - What makes them the most important?
 - Tell me more about . . .
 - How will you keep on doing/ start doing _____?
9. Who are the most important people in your life right now?
 - What makes them the most important?
 - Tell me more about . . .
10. What do you hope to be doing in the next 2 – 5 years?
 - What will you need to be able to do _____?
 - How do you imagine that you might be able to do this?
 - Who/what would be helpful for you to do this?
11. How satisfied are you with your ability to make your own decisions?
 - Rank your satisfaction on a scale of 1 – 10


- Are you more satisfied with making decisions in some areas of your life than others?
 - What areas are you most/least satisfied?



End of Life

12. Thinking about when your condition declines and you may need more care or be in a hospital/hospice, I have some questions I would like to ask you. Are you ok with this? Yes or no is fine. We can stop whenever you like.

- Do you know (or want to know) about how your (disease/condition) will affect you as your condition gets worse?
- Do you have any plans or preferences for when your health declines and you will need more care and support? Tell me more . . .
 - Some people prefer to be at home, others a hospital or hospice. Where would you like to be? Why?
- Do you have someone you can trust to make sure that you will be cared for according to your preferences and where you want to be when you are unable to speak for yourself?
 - Who is/are that person/people
 - What is it about them that allows you to trust them?
 - Are there any people that you are worried will change your plans if you are unable to speak for yourself?

Appendix F: Research Ethics Board Approval: McMaster University

① GINA in FYI
② SCAN FOR KAREN COOK
③ FILE  OK

 **RESEARCH ETHICS BOARD** 

RENEWAL FORM
Research Ethics Board Review of an Active Study

REB Project #: 11-448

Principal Investigator: Dr. Gina Browne

Project Title: The Experience of Youth with Pediatric Life Limiting Conditions Transitioning from Pediatric to Adult Services: A Case Study

Approved for Continuation

Approved conditional on changes noted in "Conditions" section below

Type of Approval:

Full Research Ethics Board

Research Ethics Board Executive


REB Approval Period: Approval period covers September 20-2012 to September 20-2013

New Enrolment Suspended

Suspended pending further review

Conditions:

The Hamilton Health Sciences/McMaster University Research Ethics Board operates in compliance with the ICH Good Clinical Practice Guidelines and the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans and Division 5 Health Canada Food and Drug Regulations.


Suzette Salama, PhD., Chair
Hamilton Health Sciences/Faculty of Health Sciences Research Ethics Board

18/09/12
Date

All Correspondence should be addressed to the REB Chair and forwarded to:
REB Coordinator, Hamilton Health Sciences
293 Wellington St. N., Suite 102, Hamilton ON L8L 8E7
Tel. 905-521-2100 Ext. 42013 Fax: 905-577-8378

Appendix G: Research Ethics Board Approval: University of British Columbia



CHILDREN'S & WOMEN'S HEALTH CENTRE OF BRITISH COLUMBIA
 AN AGENCY OF THE PROVINCIAL HEALTH SERVICES AUTHORITY
 UBC C&W Research Ethics Board
 A2-136, 950 West 28th Avenue
 Vancouver, BC V6Z 4H4
 Tel: (604) 875-3103 Fax: (604) 875-2496
 Email: cwreb@cw.bc.ca
 Website: http://www.cfn.ca/research_support > Research Ethics

ETHICS CERTIFICATE OF DELEGATED APPROVAL: RENEWAL

PRINCIPAL INVESTIGATOR: Harold Siden	DEPARTMENT: UBC/Applied Science/Nursing	UBC C&W NUMBER: H11-02388
INSTITUTION(S) WHERE RESEARCH WILL BE CARRIED OUT:		
Institution		Site
N/A		N/A
Other locations where the research will be conducted: Subject's home or their own choice of meeting place Canuck Place Children's Hospice for a review of participants' health records.		
CO-INVESTIGATOR(S): Karen A Cook		
SPONSORING AGENCIES: - Canadian Health Services Research Foundation - McMaster University		
PROJECT TITLE: The Transition of Youth from Pediatric Palliative Care to Adult Services: A Case Study (July 2012)		
REMINDER: The current UBC Children's and Women's approval for this study expires: November 13, 2013		
APPROVAL DATE: November 13, 2012		
CERTIFICATION: In respect of clinical trials: 1. The membership of this Research Ethics Board complies with the membership requirements for Research Ethics Boards defined in Division 5 of the Food and Drug Regulations. 2. The Research Ethics Board carries out its functions in a manner consistent with Good Clinical Practices. 3. This Research Ethics Board has reviewed and approved the clinical trial protocol and informed consent form for the trial which is to be conducted by the qualified investigator named above at the specified clinical trial site. This approval and the views of this Research Ethics Board have been documented in writing.		
The Chair of the UBC Children's and Women's Research Ethics Board has reviewed the documentation for the above named project. The research study, as presented in the documentation, was found to be acceptable on ethical grounds for research involving human subjects and was approved for renewal by the UBC Children's and Women's Research Ethics Board.		
<i>Approved by one of:</i>		
Dr. Marc Levine, Chair Dr. Caron Strahlendorf, Associate Chair		