PATIENT ENGAGEMENT IN AUTISM RESEARCH

Facilitators and Barriers Contributing to Patient Engagement in Autistic Children's Research

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TITLE: Facilitators and Barriers Contributing to Patient Engagement in Autism Children's Research

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Lay Abstract

Autistic children from certain marginalized population groups are less likely to be engaged in research. This thesis aims to examine the facilitators and barriers contributing to this phenomenon. As predicted, several child and family characteristics, socio-economic factors, and contextual research structures appear to be associated with patient engagement in autism research. Several recommendations are made for policymakers, researchers and funders on ways to prioritize autistic populations' needs, enhance patient engagement, and promote a more equitable approach to autism research.

Abstract

Background: Marginalized population groups are more likely to be under-represented in autistic children's research.

Rationale: Our understanding of the low levels of research engagement among autistic children from these marginalized groups is limited.

Objectives: (1) to examine some of the under-represented populations in autism children's research; (2) to assess the facilitators and barriers contributing to patient engagement in autistic children's research; (3) to suggest considerations for the development of a more equitable approach to autistic children's research.

Methods: A scoping review was conducted on studies published during January 2011-December 2021 in five electronic research databases by two reviewers in duplicate. English qualitative/quantitative/mixed methods studies that engaged autistic children aged 2-18 and/or their parents as research participants or in the process of patient-oriented research were included.

Results: Some of the under-represented marginalized autistic children populations identified from the 21 included studies were: (1) those living in developing/under-developed countries, (2) those who received autism services from centres that do not collaborate with researchers, (3) families of ethnic minority in Western countries, (4) autistic children who received late diagnosis, (5) families whose first language is not English, (6) male parents of autistic children, (7) female autistic children, (8) families with low household income who are not enrolled in governmental healthcare financial support program and (9) those who lack technological literacy skills. Facilitators of patient engagement were: (1) building trust-based relationships among stakeholders, (2) engaging patients throughout research development, and (3) patient engagement in research funding processes. The barriers were: (1) allocation of research funding, (2) identity conflict, (3) applicability of research evidence, and (4) social stigmatization towards autism.

Discussion: To enhance patient engagement in autistic children's research, policymakers, researchers and funders should prioritize participant's needs in all stages of the research process.

Conclusion: The diverse identities autistic children carry should be better acknowledged. An equity approach to research is needed.

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PREFACE:

This thesis contains a manuscript to be submitted to a journal for publication and is formatted as a "sandwich" thesis. The first chapter includes an introduction that gives an in-depth background of my thesis topic referencing previous literature. The second chapter is a manuscript that contains an introduction, methods, results, and discussion. The third chapter is a conclusion to the thesis.

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LIST OF ABBREVIATIONS AND SYMBOLS

ADHD	Attention deficit hyperactivity disorder
APA	American Psychiatric Association
DSM-5	Diagnostic and Statistical Manual of Mental Disorders (Fifth Version)
ASD	Autism spectrum disorder
PHAC	Public Health Agency of Canada
WHA	World Health Assembly
IQ	Intelligence quotient
PDD-NOS	Pervasive developmental disorder not otherwise specified
POR	Patient-oriented research
US	United States
UK	United Kingdom
PRISMA-ScR	Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews
SK	South Korea
SA	South Africa
CIHR	Canadian Institutes of Health Research
KT	Knowledge translation
REB	Research ethics board
HiREB	Hamilton Integrated Research Ethics Board
PHAC	Public Health Agency of Canada
TCPS2	Tri-Council Policy Statement
EUPATI	European Patients' Academy on Therapeutic Innovation Open Classroom

DECLARATION OF ACADEMIC ACHEIVEMENT:

I, ALICIA HOI YING LIU, declare this thesis work to be my own. The research question and design were co-developed by me and my supervisor, Dr. Georgiades in consultation with Dr. Lai and Dr Wong. I developed the scoping review search strategy, and it was verified by Rachel Couban, a librarian at the McMaster University Health Sciences Library. Data screening and data extraction were completed in duplicate by Anna Kata (second reviewer), research coordinator at the Offord Centre for Child Studies, and I (first reviewer). I wrote all sections of this thesis. It was revised based on the in-depth feedback from Dr. Georgiades, Dr. Lai, Dr. Wong and Dr. Deepa. I understand that my thesis will be available electronically via MacSphere.

Chapter 1: Introduction

1.1 Overview

This thesis aims to examine issues related to the under-representation of marginalized population groups in autism research, identify facilitators and barriers contributing to the research engagement of these groups, and outline considerations for the promotion of a more equitable approach to autism research.

Patient-oriented research (POR) that actively engages patients/research participants help to reduce power dynamics contributed by researchers and funders in research development. Those who carry marginalized identities have increased risk of unfavourable health outcomes, for instance, developing chronic diseases. Being diagnosed with autism can increase one's risk of facing social discrimination, the lack of awareness and understanding of autism in our society and the challenges autistic individuals face to communicate with the society from their diagnosis are some of the factors that contribute to this phenomenon. The unbalanced power relations among researchers, funders, and community organizations/members promotes the overpowered researchers and funders in making decisions in research development. POR allows the prioritization of patient's concerns, which is vital in producing accurate research evidence in supporting the community members and other stakeholders to make healthcare decision about their population. Engaging the children population (aged 18 and under) in research has been a challenge acknowledged by the academic research field. The intersectionality of being an autistic child and carrying other marginalized identities creates complicated and unique experiences of social oppression, which could make it particularly challenging for this population to engage in research.

1.2 Autism

1.2.1 Language in Discussing Autism

Autism is a neurodevelopmental disability usually diagnosed in childhood, that has a life-long impact on how a person communicates and interacts with the world around them (American Psychiatric Association [APA], 2013). Language is a powerful tool that shapes how we think and communicate. The terms we use in autism research often impact how our community perceives autism and autistic individuals. There has been an ongoing discussion with members of the autism community about their preferred language to make them feel safe and welcome when describing and referencing the condition (Sterponi et al., 2015).

A debate related to the language we use when discussing autism is whether the terminology "disorder" should be implemented. The fifth and most current version of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) described autism as "autism spectrum disorder (ASD)". However, I decided to discuss autism in a way that avoids using the terminology "disorder" as the word "disorder" entails negative connotations and promotes stigmatization (APA, 2013; Lord, 2018). One of the goals of this thesis is to promote an equity-based approach to research, hence I have chosen to adopt term "condition" when discussing autism. Our team strives to prevent stigmatization towards the autistic community.

The language used around the three severity levels of autism, based on DSM-5 classification, does not fully represent autistic individuals' rich talents, strengths, and the complex challenges they face. The DSM-5 classifies autistic individuals into three levels based on the assistance they need in daily life (see section "1.2.4 Autism Diagnosis"). The classification of "low functioning autism" or "high functioning autism" is also sometimes used in autism research when studying a specific severity level of autistic participants (Bottema-Beutel et al., 2021). Nonetheless, these terminologies are less preferred for the description of clinical presentation of autism since there is a concern that these label are misleading by lack

of acknowledging each autistic individual's strengths and challenges and contribute to the stigmatization of autism (Mottron, 2006). I encourage a more detailed description of their abilities and how their life is impacted. Consequently, in the scoping review completed for and attached to this thesis, I attempted to record all the characteristics mentioned about the autistic individuals who participated in the included research studies, rather than only using just the autism diagnosis.

There are diverse opinions for the use of identity-first language (for example, "autistic person") or person-first language (for example, "person with autism"). On one hand, there are concerns that using identity-first language has the potential for increasing stigma towards autism; on the other hand, identity-first language acknowledges that being autistic is an integral part of a person's identity. I understand that every individual on the autism spectrum might have a different preference on person-first or identity-first language. In this thesis, I have chosen to adopt the term "autistic" since it was found to be the preferred term in a large survey of autistic adults, families, friends and parents (Kenny et al., 2016). Although the term "autistic" is less preferred than "person with autism" among professionals, I chose to prioritize patients' preferences, as this thesis focuses on POR, that places patients at the centre of their research studies. I will also use the phrase "on the spectrum" or "on the (autism) spectrum" to describe autistic individuals interchangeably, which will be further discussed in section "1.2.3 Autistic Symptoms". The phrases "suffers from" and "is a victim of autism" will be avoided so as not to victimize the autistic individuals.

Lastly, another aspect of language used to understand autism is to compare how this condition impacts autistic individuals' daily living as compared to those who are not diagnosed with autism. I will avoid using the term "normal" or "normally developing" and "healthy" when referring to the comparison groups; rather I will refer to them as "neurotypical" or

"typically developing children" in this thesis. This is to show full respect to autistic individuals and prevent being offensive to them.

1.2.2 Autism Epidemiology

Autism affects 1-2% of the population around the world (APA, 2013; Elsabbagh et al., 2012). According to the 2019 Canadian Health Survey on Children and Youth, 1 in 50 (2.0%) 1 to 17 years old Canadian children and youth, across ten provinces and three territories, were diagnosed with autism (Public Health Agency of Canada [PHAC], 2022). Specifically, the prevalence of autism among males was four times higher than females, which results in an imbalanced sex ratio population towards males in community-based autism research (Beggiato et al., 2017; PHAC, 2016). However, it is believed that gender bias contributes to the underestimation and increased risk of misdiagnosis of autism in females (Haney, 2016). For instance, fewer restricted and repetitive behaviours are often observed in autistic females compared to autistic males, but the diagnostic guidelines were developed based on common symptoms among male autistic individuals (Werling & Geschwind, 2013). As a result, it can be difficult to report on gender diversity in autistic population.

In Canada, the lifespan value of caregiver time, per individual required to support a severely impacted autistic child, is approximately $5.5 million higher than that for someone without autism (Dudley & Emery, 2014). At the population level, this had already exceeded the combined annual direct medical, non-medical and productivity costs of stroke and hypertension, and this cost was predicted to rise from $268 billion in 2015 to $461 billion in 2025 (Leigh & Du, 2015). In 2014, the World Health Organization adopted a resolution of "comprehensive and coordinated efforts for the management of [ASD]", at the sixty-seventh World Health Assembly (WHA) that urges strengthening of national capacities to address autism (World Health Assembly [WHA], 2014). A commitment was made to address the needs of individuals, families and societies affected by autism by safeguarding the autistic

community from social stigmatization, isolation, and discrimination (WHA, 2014). Autism is an emerging public health concern not only locally in Canada, but also in many countries around the world.

1.2.3 Autistic Symptoms

Autism is a highly heterogeneous condition. A wide variation in the type and severity of symptoms are observed among autistic individuals, but to date, no unifying genetic or environmental factors have been identified to account for the condition (Garel & Garel, 2019). These differences among autistic individuals involve a mix of strengths and challenges in their life. For instance, some autistic individuals show no functional language skills while others demonstrate average or superior intelligence quotient (IQ). This broad spectrum of autism phenotypes is emphasized by the word "spectrum" in autism spectrum condition.

Autism symptoms affect autistic individuals' daily life (Seltzer et al., 2003). Some of the most commonly observed signs of autism include challenges in social communication and interaction, avoiding eye contact, engaging in repetitive patterns of movements, following a strict daily routine and becoming anxious or upset by minor changes, and experiencing sensory hyper-sensitivity (APA, 2013; Seltzer et al., 2003). The symptoms can affect an individual throughout the life course (Posar & Visconti, 2019).

1.2.4 Autism Diagnosis

An autism diagnosis is made by qualified professionals who have undergone special training (Hayes et al., 2022). Some of these professionals include family physicians, paediatricians, developmental paediatricians, psychiatrists, and psychologists (Hayes et al., 2022). Currently, there is no medical test (for example, blood test) for autism diagnosis (Dover & Couteur, 2007; Hayes et al., 2022). An autism diagnosis is usually made based on the child's developmental history and behaviour using an assessment tool (Dover & Couteur, 2007). One of the most commonly used diagnostic manual is the Diagnostic and Statistical Manual of

Mental Disorder (DSM). Based on the fifth and most current version of the DSM (DSM-5), autism is subdivided into three levels of severity (Appendix), this is determined based on the level of support an autistic individual needs in the area of social communication and restricted/repetitive behaviour (APA, 2013).

The revised DSM published in 2013,, made several changes in autism diagnostic criteria and classifications (APA, 2013). People diagnosed with autistic disorder, Asperger's syndrome and pervasive developmental disorder not otherwise specified (PDD-NOS) are now classified as a form of autism (APA, 1994; APA, 2013). In this thesis, I mean to include Asperger's syndrome and PDD-NOS when referring to autism.

There are challenges in accurate and timely autism diagnoses. As mentioned, unique individuals on the spectrum are impacted very differently, with a wide range of symptoms and severity; this makes diagnosing autism to be difficult for health professionals (Hayes et al., 2022). The challenges to diagnose autism do not only delay those on the spectrum to receive support in a timely manner, but it can also impact the design of autism research, specifically it can make recruiting patient samples of the autism population become complicated.

1.3 Patient Engagement in Patient-Oriented Research

Patient-oriented research (POR) is built upon the principal of "nothing about me without me" (Sacristán, 2013). In POR, gathering input from patients, at the centre of the discussion of their health, is important when deciding on how the research will be conducted and used for (Sacristán et al., 2016). This is as opposed to restricting patients' contributions in research as merely research participants being studied by the researchers. The quality of community-engaged research is influenced by the extent of active collaboration between researchers and community members (Sacristán et al., 2016). Patients' and clinicians' research priorities and values can be different (Crowe et al., 2015). POR listens to the voices of those

with lived experience by addressing the outcomes deemed important by patients (Baumgardner, 2019). In order to allow patients to meaningfully engage as part of a POR, it is required to level the power dynamics in order to prioritize patients' value when making decisions in the planning and conducting of research. This allows shared decision making among the patients, academic researchers, and funding organizations. In good practice, stakeholder engagement should take place throughout all stages of a patient-oriented research, including the preparatory phase, execution phase, evidence synthesis, data interpretation, and findings dissemination (Selker et al., 2010). POR equips the patient population to make decisions using applicable research evidence that is tailored to address the questions of relevance to them and support the healthcare providers and policymakers to design and deliver health services that meets their needs (Sacristán et al., 2016).

Power dynamics are the unequally shared capacity that exists by virtue of a party's title or social position. This is especially influential in how decisions are made; the point of view or concern of those with greater power tends to weigh more heavily and is more likely to be considered when making a decision. In the field of conducting academic research, the power relations among research participants, community organizations, researchers and funding committees impact the kind of research produced and healthcare decisions made (Baum, 2006; Riley, 2003). Most of the current research is dictated by funding organizations' preferences and academic researchers familiar with how to conduct research, rather than conducting POR (Boivin et al., 2018). For instance, one of the best-known comparison research studies investigated the priorities in the assessment of treatments for osteoarthritis of the knee. This resulted in marked differences between the researchers' preferred drug trials versus the non-drug treatment research prioritised by patients, carers and clinicians (Tallon et al., 2000). The same results were confirmed more recently by studying general treatment research priorities (Crowe et al., 2015).

There are benefits and risks for research participants and researchers to engage collaboratively in research. In addition to benefiting from being empowered to generate research findings that can support their health decision-making progress, research participants will also increase their knowledge in the research topic (Castillo et al., 2012). Nevertheless, there are risks to participating in POR. There is risk for research participants to share their identifiable information. Also, the additional time and cost required for participating in most of the POR might not have direct benefits on the participants. When conducting research, the benefits and risks must be balanced. Therefore, it is challenging to engage participants in POR.

In the context of autism, it is particularly important to have an emphasis of patient engagement by conducting autistic POR. A clear disparity between autism research funding patterns/research priorities and autistic stakeholders' research concerns/priorities is reported among different autistic communities (Pellicano et al., 2014; Russell et al., 2018). A call has been made for more emphasis on applied science research that can address more immediate concerns of autistic individuals, compared to large amounts of basic science/pre-clinical research previously funded in the field. In a survey conducted in the United States (US) with 6004 respondents, autistic individuals rated the investigation of the scientific causes of autism just below the neutral rating, while family members, researchers and clinician/educator all rated this topic as moderately impactful (Frazier et al., 2018). Similar differences between autistic individuals and researchers' priorities were observed in the United Kingdom (UK) (Pellicano, 2014). Even with the large body of autism laboratory research conducted, active patient engagement to contribute to experimental development and dissemination of results is scarce (Johnston, 2021).

1.4 Marginalized Populations

Marginalized populations are groups and/or communities that experience discrimination and exclusion from society. Society provides few opportunities for the engagement of disadvantaged populations when making social, political and economic decisions. The unequal power relationships among different social groups contributes to marginalization. Some of the marginalized autistic populations recognized from previous literature include, but are not limited to, groups disadvantaged due to race, sex, gender, sexual orientation, age, culture, language, and/or immigration status (Seng et al., 2012). Marginalized groups have high risk of poor health outcomes (Farrell, 2005). Minority groups are more likely to develop chronic diseases. For instance, Black Canadians are more likely to develop hypertension or diabetes compared to White Canadians (Gagné & Veenstra, 2017). We should pay additional attention to how intersectionality impacts marginalized groups. Overlapping marginalized social identities creates complex and distinct experiences of discrimination and oppression (Crenshaw, 1991). In this example, the prevalence of developing hypertension or diabetes were higher among Black Canadian women compared to White Canadian women (Gagné & Veenstra, 2017).

Being diagnosed with autism is an identity that increases individuals' chances of experiencing social marginalization, and they are thus more likely to face discrimination (O'Connor, 2020). The nature of autism symptoms impacts one's social communication ability that often makes autistic individuals more socially isolated. An autism diagnosis is just one of the many identities that autistic individuals carry. It does not fully represent who they are as a person. It is important to acknowledge that each of the community members engaged in autism research development also carry other identities. Autistic individuals who hold marginalized identities are more likely to experience delayed access to diagnostic and early intervention autism services (Bishop-Fitzpatrick & Kind, 2017). When an autistic individual carries more

than one marginalized identity, they often encounter greater disparities in health status and life expectancy than the privileged individuals who hold more power in the society (Bishop-Fitzpatrick & Kind, 2017). As a result, addressing the marginalized populations in autism research is a priority.

A good practice of patient engagement in POR should engage the true representation of the targeted population as research participants. An inclusive research environment and diverse approaches to engagement is needed in order to engage people with different identities in the community to engage in research. When the research sample is only a partial representation of the population, the biased research may generate information that does not represent the experiences of all individuals with different identities of the group of interest. This impacts how healthcare decisions are made and how resources are allocated (Baah, 2019). Therefore, it is important to study how participants are recruited in POR.

Marginalized populations are likely to experience increased risks from participating in POR. Particularly, this might expose their marginalized identities and increase their risk in facing additional social discrimination and oppression. Research ethics boards are therefore more likely to request for the research studies engaging these marginalized populations to put more emphasis in ensuring the risks are appropriately addressed to protect them. However, the power imbalance between research ethics boards and community members could mean shifting the research focus, moving away from what patients value. There are also risks for researchers who study a sensitive topic that engages marginalized populations, especially those that they are not a part of. This could make researchers feel reluctant to conduct research of marginalized populations. Therefore, it makes it even more challenging to engage marginalized populations in research.

There are concerns that the processes and strategies that were used to engage participants in POR systematically under-represented marginalized populations from

meaningful engagement in autism research development (Fletcher-Watson et al., 2019; McCoy, 2020; West et al., 2016; Zwicker, 2014). For example, there is a lack of autism research engaging black and African families (Shaia et al., 2020). This reveals that there is a non-inclusive research engagement environment in the field. Limited engagement of marginalized populations results in biased research with inaccurate information about the autism population. By studying how sampling was done in autism research studies, we can identify who are the autistic individuals included as research participants, which could provide insights on some of the under-represented marginalized population. Addressing the issue of marginalized populations in autism research helps to ensure that the POR conducted by the autistic population includes all the communities that were intended to address.

1.5 Patient-Oriented Research Engaging Children Population

POR that studies children (age 18 or under) is limited in understanding the children's experiences through patient engagement due to complex methodological and ethical considerations (Schmidt & Paris, 1984). There is a lack of systematic and explicit research agenda to engage paediatric chronic disease patients in research (Odgers et al., 2018).

The "why am I always being researched?" guide developed by Chicago Beyond is one of the frameworks that addresses the concerns in engaging paediatric population in research. The guide is built based on the voices of community leaders and organizations, instead of researchers or funders (Chicago Beyond, 2018). Based on lived experiences, community members summarized seven inequalities that hinder them from engaging in research (Chicago Beyond, 2018). This guide is developed aiming to improve youth equity, specifically promoting for more equitable access and opportunity to young people in Chicago (Chicago Beyond, 2018). "Access", "information", "validity", "ownership", "value", "accountability" and "authorship" are the seven inequalities that are held in place by the power dynamics among

community organizations, researchers and funders (Chicago Beyond, 2018). "Access" refers to the challenge for members of the community to access creating knowledge about their communities; "information" inequality prevents the community from being engaged in research as they do not have enough information to contribute their wisdom to the research development; the "validity" of information shared by the community is often less prioritized than institutional data; community members contributing to research do not have the "ownership" to data produced in the study; the "value" of research for the community is not always clear to the community members; the community "accounts" for the greatest risks as the researchers or funders do not always take the responsibility when the study design does not work; the "authorship" of community organizations are restricted and their voices are muted (Chicago Beyond, 2018). By addressing these seven inequalities identified by Chicago Beyond and the community leaders and organizations with which they work, it is expected that it will help promote equity in research development (Chicago Beyond, 2018). The promotion of equity-based approaches in research is important when uncovering knowledge that targets additional marginalized identities. This helps to balance the power among these parties when conducting research with autistic children. By enhancing patient engagement, the autism POR produced can better support this marginalized population.

The risks of engaging children as research participants require special attention. There is less data on clinical research engaging children. The first guidance by the Royal College of Paediatrics and Child Health in relation to research engaging children was published in 1980 (British Paediatric Association, 1980). Prior to this time, little clinical research engaged children (Modi et al., 2014). This still remains as a topic of discussion for research ethics boards in Canada (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Since there is less data available to reference when conducting research engaging the children population, it increases the uncertainties and risks of engaging this population. This results in

an extra cautious attitude for research ethics boards to approve pediatric research, which could magnify the power imbalance between research and ethics. Ultimately, creating the challenge to engage the children population in POR and reducing the resources available for this kind of research, leads to an unhealthy vicious cycle.

Paediatric researchers are obligated to protect and promote children's rights when engaging them as research participants. This includes ensuring that children have the right to express their views about the research process itself and give informed consent (Bell, 2008). However, engaging young children in research can be difficult. Although language development peaks among typically developing toddlers and pre-schoolers (Foster-Cohen, 2013), this children population might not have developed the proficient language skills to fully express themselves verbally, and therefore makes involving toddlers and pre-schoolers in research extraordinarily hard. In autism research, autistic children who are minimally verbal or with less developed non-verbal skills might also make it challenging in understanding their views on research priorities (McCoy, 2020; Tesfaye et al., 2019). The lack of language tools for autistic children to speak up for themselves exaggerates the differences in power capacity of other stakeholders when conducting autism research. Therefore, additional effort to design novel strategies, including interpretation of non-verbal communication responses, use of Photo-elicitation research technique and flexible implementation of research protocol, can be needed to engage autistic children (Courchesne et al., 2021; Tesfaye et al., 2019). This is a specific ethical challenge to tackle when engaging children, in particular autistic children, in the planning and conducting of POR.

Some current paediatric research depends on recruiting stakeholders, including caregivers, siblings, teachers, and knowledge users, as study participants. Caregivers are one of the commonly studied stakeholders in autism research. Caregiver is the person who takes the main responsibility in taking care of the child, most commonly this refers to the child's

mother or father. Given the challenges in engaging autistic children in research, it is often assumed that the caregiver with whom an autistic child spends a lot of their time growing up is the person who can help us to understand the child. However, it is important to be aware that caregivers can only try their best to understand the youngsters. Most of the time it relies on the caregiver's observational skills on the child's preferences based on their non-verbal cues (McCoy, 2020; Tesfaye et al., 2019). It should not be always assumed that the caregiver's feedback in research priorities is true to the autistic children and family's personal feeling and thoughts (McCoy, 2020; Tesfaye et al., 2019). Studies that used autistic children's first-voice where ethically possible is important to conducting inclusive POR (Tesfaye et al., 2019).

Given the factors mentioned above, our primary hypothesis is the different marginalized identities that autistic children carry makes it more challenging for this population to be engaged in research. Our secondary hypothesis is that autistic children's first voice is under-represented in research. Addressing this misrepresented autistic community is vital to provide opportunities of empowerment and enhancing communication while ensuring that the decisions made on their health is based on their priorities (Danker et al., 2017). Since autism is reliably diagnosed at age two onwards, this thesis will study marginalized autistic children aged 2 to 18 (Centers for Disease Control and Prevention, 2020). The research question of this thesis is: Who and how were the routinely marginalized autistic children aged 2 to 18 engaged in academic research? A secondary analysis of autism research engaging children's first-voice versus those relying on the engagement of caregivers will be made.

1.6 Public Health Implications

This study addresses the vulnerability of marginalized autistic subpopulations and appreciates the diverse identities each autistic individual carries by promoting an equity-based approach to research. It aims to recognize some of the marginalized populations that are under-

represented in autism research development and begins to address the different facilitators and barriers that affect how marginalized patients are engaged in autistic children's research. It is expected that this study will improve how autism research is conducted. Through making suggestions on considerations that should be addressed when conducting autism research, it will also help create greater impact of health decisions to help provide the right support that is needed for all Autistics.

1.7 Thesis objectives

1. To identify the marginalized autistic population groups that have been included in autistic children's research, and to suggest some of the under-represented autistic population groups in research.
2. To assess the facilitators and barriers contributing to patient engagement in autistic children's research.
3. To suggest considerations for the development of a more equitable approach to research on autistic children.

References

American Psychiatric Association. (1994). *DSM-IV: Diagnostic and Statistical Manual of Mental Disorders*. https://doi.org/10.1001/jama.1994.03520100096046

American Psychiatric Association. (2013). *DSM-V: Diagnostic and Statistical Manual of Mental Disorders*. http://archive.org/details/diagnosticstatis0005unse

Baah, F. O., Teitelman, A. M., & Riegel, B. (2019). Marginalization: Conceptualizing patient vulnerabilities in the framework of social determinants of health-An integrative review. *Nursing Inquiry*, *26*(1), e12268. https://doi.org/10.1111/nin.12268

Baum, F., MacDougall, C., & Smith, D. (2006). Participatory action research. *Journal of Epidemiology and Community Health*, *60*(10), 854–857. https://doi.org/10.1136/jech.2004.028662

Baumgardner, D. J. (2019). Defining Patient-Oriented Research for the Average Person (and Potential Research Partner). *Journal of Patient-Centered Research and Reviews*, *6*(1), 4–6. https://doi.org/10.17294/2330-0698.1697

Beggiato, A., Peyre, H., Maruani, A., Scheid, I., Rastam, M., Amsellem, F., Gillberg, C. I., Leboyer, M., Bourgeron, T., Gillberg, C., & Delorme, R. (2017). Gender differences in autism spectrum disorders: Divergence among specific core symptoms. *Autism Research*, *10*(4), 680–689. https://doi.org/10.1002/aur.1715

Bell, N. (2008). Ethics in child research: Rights, reason and responsibilities. *Children's Geographies*, *6*(1), 7–20. https://doi.org/10.1080/14733280701791827

Bishop-Fitzpatrick, L., & Kind, A. J. H. (2017). A Scoping Review of Health Disparities in Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *47*(11), 3380–3391. https://doi.org/10.1007/s10803-017-3251-9

Boivin, A., Richards, T., Forsythe, L., Grégoire, A., L'Espérance, A., Abelson, J., & Carman, K. L. (2018). Evaluating patient and public involvement in research. *BMJ*, *363*, k5147. https://doi.org/10.1136/bmj.k5147

Bottema-Beutel, K., Kapp, S. K., Lester, J. N., Sasson, N. J., & Hand, B. N. (2021). Avoiding Ableist Language: Suggestions for Autism Researchers. *Autism in Adulthood*, *3*(1), 18–29. https://doi.org/10.1089/aut.2020.0014

British Paediatric Association. (1980). Guidelines to aid ethical committees considering research involving children. *Archives of Disease in Childhood*, *55*(1), 75–77.

Castillo, A. G., Jandorf, L., Thélémaque, L. D., King, S., & Duhamel, K. (2012). Reported Benefits of Participation in a Research Study. *Journal of Community Health*, *37*(1), 59–64. https://doi.org/10.1007/s10900-011-9416-0

Centers for Disease Control and Prevention. (2020, March 13). *Screening and Diagnosis | Autism Spectrum Disorder (ASD) | NCBDDD*. Centers for Disease Control and Prevention. https://www.cdc.gov/ncbddd/autism/screening.html

Chicago Beyond. (2018). *Why am I always being researched?* (p. 111). https://chicagobeyond.org/wp-content/uploads/2019/05/ChicagoBeyond_2019Guidebook.pdf

Courchesne, V., Tesfaye, R., Mirenda, P., Nicholas, D., Mitchell, W., Singh, I., Zwaigenbaum, L., & Elsabbagh, M. (2021). Autism Voices: A novel method to access first-person perspective of autistic youth. *Autism*, 13623613211042128. https://doi.org/10.1177/13623613211042128

Crenshaw, K. (1991). Mapping the Margins: Intersectionality, Identity Politics, and Violence against Women of Color. *Stanford Law Review*, *43*(6), 1241–1299. https://doi.org/10.2307/1229039

Crowe, S., Fenton, M., Hall, M., Cowan, K., & Chalmers, I. (2015). Patients', clinicians' and the research communities' priorities for treatment research: There is an important mismatch. *Research Involvement and Engagement, 1(1),* 2. https://doi.org/10.1186/s40900-015-0003-x

Danker, J., Strnadová, I., & Cumming, T. M. (2017). Engaging Students With Autism Spectrum Disorder in Research Through Participant-Driven Photo-Elicitation Research Technique. *Australasian Journal of Special Education*, *41*(1), 35–50. https://doi.org/10.1017/jse.2016.7

Dover, C. J., & Couteur, A. L. (2007). How to diagnose autism. *Archives of Disease in Childhood*, *92*(6), 540–545. https://doi.org/10.1136/adc.2005.086280

Dudley, C., & Emery, J. C. H. (2014). *The Value of Caregiver Time: Costs of Support and Care for Individuals Living with Autism Spectrum Disorder* (SSRN Scholarly Paper ID 2379633). Social Science Research Network. https://doi.org/10.2139/ssrn.2379633

Elsabbagh, M., Divan, G., Koh, Y.-J., Kim, Y. S., Kauchali, S., Marcín, C., Montiel-Nava, C., Patel, V., Paula, C. S., Wang, C., Yasamy, M. T., & Fombonne, E. (2012). Global Prevalence of Autism and Other Pervasive Developmental Disorders. *Autism Research*, *5*(3), 160–179. https://doi.org/10.1002/aur.239

Farrell, Ann (Ed.) (2005) *Ethical Research with Children.* Open University Press/ McGraw-Hill Education, UK, Buckinghamshire, Milton Keynes.

Foster-Cohen, Susan H. (2013). *An Introduction to Child Language Development*. Routledge. https://doi.org/10.4324/9781315844893

Frazier, T. W., Dawson, G., Murray, D., Shih, A., Sachs, J. S., & Geiger, A. (2018). Brief Report: A Survey of Autism Research Priorities Across a Diverse Community of Stakeholders. *Journal of Autism and Developmental Disorders*,

48(11), 3965–3971. https://doi.org/10.1007/s10803-018-3642-6

Fletcher-Watson, S., Adams, J., Brook, K., Charman, T., Crane, L., Cusack, J., Leekam, S., Milton, D., Parr, J. R., & Pellicano, E. (2019). Making the future together: Shaping autism research through meaningful participation. *Autism*, *23*(4), 943–953. https://doi.org/10.1177/1362361318786721

Gagné, T., & Veenstra, G. (2017). Inequalities in Hypertension and Diabetes in Canada: Intersections between Racial Identity, Gender, and Income. *Ethnicity & Disease*, *27*(4), 371–378. https://doi.org/10.18865/ed.27.4.371

Garel, N., & Garel, P. (2019). Diagnosis of Autism Spectrum Disorder in Adolescents with Complex Clinical Presentations: A Montreal Case Series. *Adolescent Psychiatry*, *9*(1), 33–43.

Haney, J. L. (2016). Autism, females, and the DSM-5: Gender bias in autism diagnosis. *Social Work in Mental Health*, *14*(4), 396–407. https://doi.org/10.1080/15332985.2015.1031858

Hayes, J., Ford, T., McCabe, R., & Russell, G. (2022). Autism diagnosis as a social process. *Autism*, *26*(2), 488–498. https://doi.org/10.1177/13623613211030392

Interagency Advisory Panel on Research Ethics Government of Canada. (2019, April 1). *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans – TCPS 2 (2018) – Chapter 4: Fairness and Equity in Research Participation*. https://ethics.gc.ca/eng/tcps2-eptc2_2018_chapter4-chapitre4.html#6

Johnston, J. N., Ridgway, L., Cary-Barnard, S., Allen, J., Sanchez-Lafuente, C. L., Reive, B., Kalynchuk, L. E., & Caruncho, H. J. (2021). Patient oriented research in mental health: Matching laboratory to life and beyond in Canada. *Research Involvement and Engagement*, *7*(1), 21. https://doi.org/10.1186/s40900-021-00266-1

Kenny, L., Hattersley, C., Molins, B., Buckley, C., Povey, C., & Pellicano, E. (2016). Which terms should be used to describe autism? Perspectives from the UK autism community. *Autism*, *20*(4), 442–462. https://doi.org/10.1177/1362361315588200

Leigh, J. P., & Du, J. (2015). Brief Report: Forecasting the Economic Burden of Autism in 2015 and 2025 in the United States. *Journal of Autism and Developmental Disorders*, *45*(12), 4135–4139. https://doi.org/10.1007/s10803-015-2521-7

Lord, C., Elsabbagh, M., Baird, G., & Veenstra-Vanderweele, J. (2018). Autism spectrum disorder. *The Lancet*, *392*(10146), 508–520. https://doi.org/10.1016/S0140-6736(18)31129-2

McCoy, M. S., Liu, E. Y., Lutz, A. S. F., & Sisti, D. (2020). Ethical Advocacy Across the Autism Spectrum: Beyond Partial Representation. *The American Journal of Bioethics: AJOB*, *20*(4), 13–24. https://doi.org/10.1080/15265161.2020.1730482

Modi, N., Vohra, J., Preston, J., Elliott, C., Hoff, W. V., Coad, J., Gibson, F., Partridge, L., Brierley, J., Larcher, V., & Greenough, A. (2014). Guidance on clinical research involving infants, children and young people: An update for researchers and research ethics committees. *Archives of Disease in Childhood*, *99*(10), 887–891. https://doi.org/10.1136/archdischild-2014-306444

Mottron, L., Dawson, M., Soulières, I., Hubert, B., & Burack, J. (2006). Enhanced Perceptual Functioning in Autism: An Update, and Eight Principles of Autistic Perception. *Journal of Autism and Developmental Disorders*, *36*(1), 27–43. https://doi.org/10.1007/s10803-005-0040-7

O'Connor, C., Burke, J., & Rooney, B. (2020). Diagnostic Disclosure and Social Marginalisation of Adults with ASD: Is There a Relationship and What

Mediates It? *Journal of Autism and Developmental Disorders*, *50*(9), 3367–3379. https://doi.org/10.1007/s10803-019-04239-y

Odgers, H. L., Tong, A., Lopez-Vargas, P., Davidson, A., Jaffe, A., McKenzie, A., Pinkerton, R., Wake, M., Richmond, P., Crowe, S., Caldwell, P. H. Y., Hill, S., Couper, J., Haddad, S., Kassai, B., & Craig, J. C. (2018). Research priority setting in childhood chronic disease: A systematic review. *Archives of Disease in Childhood*, *103*(10), 942–951. https://doi.org/10.1136/archdischild-2017-314631

Pellicano, E., Dinsmore, A., & Charman, T. (2014). What should autism research focus upon? Community views and priorities from the United Kingdom. *Autism*, *18*(7), 756–770. https://doi.org/10.1177/1362361314529627

Posar, A., & Visconti, P. (2019). Long-term outcome of autism spectrum disorder. *Turkish Archives of Pediatrics/Türk Pediatri Arşivi*, *54*(4), 207–212. https://doi.org/10.14744/TurkPediatriArs.2019.16768

Public Health Agency of Canada. (2016, January 18). *Signs and symptoms of autism spectrum disorder (ASD)* [Education and awareness]. https://www.canada.ca/en/public-health/services/diseases/autism-spectrum-disorder-asd/signs-symptoms-autism-spectrum-disorder-asd.html

Public Health Agency of Canada. (2022, February 3). *Autism spectrum disorder: Highlights from the 2019 Canadian health survey on children and youth* [Research]. https://www.canada.ca/en/public-health/services/publications/diseases-conditions/autism-spectrum-disorder-canadian-health-survey-children-youth-2019.html

Riley, S., Schouten, W., & Cahill, S. (2003). Exploring the Dynamics of Subjectivity

and Power Between Researcher and Researched. *Forum Qualitative Sozialforschung / Forum: Qualitative Social Research*, *4*(2), Article 2. https://doi.org/10.17169/fqs-4.2.713

Russell, G., Starr, S., Elphick, C., Rodogno, R., & Singh, I. (2018). Selective patient and public involvement: The promise and perils of pharmaceutical intervention for autism. *Health Expectations*, *21*(2), 466–473. https://doi.org/10.1111/hex.12637

Sacristán, J. A. (2013). Patient-centered medicine and patient-oriented research: Improving health outcomes for individual patients. *BMC Medical Informatics and Decision Making*, *13*, 6. https://doi.org/10.1186/1472-6947-13-6

Sacristán, J. A., Aguarón, A., Avendaño-Solá, C., Garrido, P., Carrión, J., Gutiérrez, A., Kroes, R., & Flores, A. (2016). Patient involvement in clinical research: Why, when, and how. *Patient Preference and Adherence*, *10*, 631–640. https://doi.org/10.2147/PPA.S104259

Schmidt, C. R., & Paris, S. G. (1984). The development of verbal communicative skills in children. *Advances in Child Development and Behavior*, *18*, 1–47. https://doi.org/10.1016/s0065-2407(08)60368-7

Selker, H. P., Leslie, L. K., Wasser, J. S., Plaut, A. G., Wilson, I. B., & Griffith, J. L. (2010). Tufts CTSI: Comparative effectiveness research as a conceptual framework for a focus on impact. *Clinical and Translational Science*, *3*(2), 56–58. https://doi.org/10.1111/j.1752-8062.2010.00184.x

Seltzer, M. M., Krauss, M. W., Shattuck, P. T., Orsmond, G., Swe, A., & Lord, C. (2003). The Symptoms of Autism Spectrum Disorders in Adolescence and Adulthood. *Journal of Autism and Developmental Disorders*, *33*(6), 565–581. https://doi.org/10.1023/B:JADD.0000005995.02453.0b

Seng, J. S., Lopez, W. D., Sperlich, M., Hamama, L., & Meldrum, C. D. R. (2012).

Marginalized identities, discrimination burden, and mental health: Empirical exploration of an interpersonal-level approach to modeling intersectionality. *Social Science & Medicine (1982)*, *75*(12), 2437–2445. https://doi.org/10.1016/j.socscimed.2012.09.023

Shaia, W. E., Nichols, H. M., Dababnah, S., Campion, K., & Garbarino, N. (2020). Brief Report: Participation of Black and African-American Families in Autism Research. Journal of Autism and Developmental Disorders, 50(5), 1841–1846. https://doi.org/10.1007/s10803-019-03926-0

Sterponi, L., de Kirby, K., & Shankey, J. (2015). Rethinking language in autism. *Autism*, *19*(5), 517–526. https://doi.org/10.1177/1362361314537125

Tallon, D., Chard, J., & Dieppe, P. (2000). Relation between agendas of the research community and the research consumer. *Lancet (London, England)*, *355*(9220), 2037–2040. https://doi.org/10.1016/S0140-6736(00)02351-5

Tesfaye, R., Courchesne, V., Yusuf, A., Savion-Lemieux, T., Singh, I., Shikako-Thomas, K., Mirenda, P., Waddell, C., Smith, I. M., Nicholas, D., Szatmari, P., Bennett, T., Duku, E., Georgiades, S., Kerns, C., Vaillancourt, T., Zaidman-Zait, A., Zwaigenbaum, L., & Elsabbagh, M. (2019). Assuming ability of youth with autism: Synthesis of methods capturing the first-person perspectives of children and youth with disabilities. *Autism: The International Journal of Research and Practice*, *23*(8), 1882–1896. https://doi.org/10.1177/1362361319831487

Werling, D. M., & Geschwind, D. H. (2013). Sex differences in autism spectrum disorders. *Current Opinion in Neurology*, *26*(2), 146–153. https://doi.org/10.1097/WCO.0b013e32835ee548

West, E. A., Travers, J. C., Kemper, T. D., Liberty, L. M., Cote, D. L., McCollow, M.

M., & Stansberry Brusnahan, L. L. (2016). Racial and Ethnic Diversity of Participants in Research Supporting Evidence-Based Practices for Learners With Autism Spectrum Disorder. *The Journal of Special Education*, *50*(3), 151–163. https://doi.org/10.1177/0022466916632495c

World Health Assembly, Sixty-Seventh (2014). Comprehensive and coordinated efforts for the management of autism spectrum disorders. WHA67.8.

Zwicker, J. D., & Emery, J. C. H. (2014). Autism research funding allocation: Can economics tell us if we have got it right? *Autism Research: Official Journal of the International Society for Autism Research*, *7*(6), 704–711. https://doi.org/10.1002/aur.1423

1.9 Appendix

Autism Severity Levels (APA, 2013)

Severity level	Social communication	Restricted, repetitive behaviours
Level 3 "Requiring very substantial support"	Severe deficits in verbal and nonverbal social communication skills cause severe impairments in functioning, very limited initiation of social interactions, and minimal response to social overtures from others. For example, a person with few words of intelligible speech who rarely initiates interaction and, when he or she does, makes unusual approaches to meet needs only and responds to only very direct social approaches.	Inflexibility of behaviour, extreme difficulty coping with change, or other restricted/repetitive behaviours markedly interfere with functioning in all spheres. Great distress/difficulty changing focus or action.
Level 2 "Requiring substantial support"	Marked deficits in verbal and nonverbal social communication skills; social impairments apparent even with supports in place; limited initiation of social interactions; and reduced or abnormal responses to social overtures from others. For example, a person who speaks simple sentences, whose interaction is limited to narrow special interests, and who has markedly odd nonverbal communication.	Inflexibility of behaviour, difficulty coping with change, or other restricted/repetitive behaviours appear frequently enough to be obvious to the casual observer and interfere with functioning in a variety of contexts. Distress and/or difficulty changing focus or action.
Level 1 "Requiring support"	Without supports in place, deficits in social communication cause noticeable impairments. Difficulty initiating social interactions, and clear examples of atypical or unsuccessful responses to social overtures of others. May appear to have decreased interest in social interactions. For example, a person who is able to speak in full sentences and engages in communication but whose-to-and-fro conversation with other fails, and whose attempt to make friends are odd and typically unsuccessful.	Inflexibility of behaviour causes significant interference with functioning in one or more contexts. Difficulty switching between activities. Problems of organization and planning hamper independence.

Chapter 2: Manuscript

Candidate's role in the manuscript: I contributed to defining the research objectives, critically reviewing the literature, developing search strategies for screening, screening studies and extracting data as first reviewer, interpretating the study findings, and writing the draft of the manuscript.

Facilitators and Barriers Contributing to Patient Engagement in Autistic Children's Research: A Scoping Review

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2.1 Abstract

Background: Research engagement is likely to be limited among autistic children with intersected marginalized identities. Under-represented marginalized populations in autistic children's research were observed.

Objective: (1) to examine who are some of the under-represented population groups in autism research; (2) to assess the facilitators and barriers contributing to patient engagement in autistic children's research; (3) to suggest considerations that should be addressed for a more equitable approach to research on autistic children.

Methods: A scoping review of autism studies that had a focus in patient engagement and published during January 2011-December 2021 in five research databases were screened by two reviewers in duplicate. English qualitative/quantitative/mixed methods studies that actively engaged autistic children aged 2-18 and/or their parents were included. Study characteristics, participant characteristics, strengths and limitations in patient engagement, and the facilitators and barriers contributing to participants' engagement in autism-related research were extracted.

Results: 21 studies were included, all of which engaged parents while none engaged solely autistic children. Some of the identified under-represented marginalized autistic children populations were: (1) those living in developing/under-developed countries, (2) those who received autism services from treatment centres that do not have collaboration with researchers, (3) families of ethnic minority in Western countries, (4) autistic children who received late diagnosis, (5) families whose first language is not English, (6) male parents of autistic children, (7) female autistic children, (8) families with low household income who are not enrolled in governmental healthcare financial support program and (9) those who do not have access to technological devices/lack technological literacy skills. Facilitators of patient engagement were: (1) building longstanding, trust-based relationships among stakeholders, (2) engaging patients throughout research development, and (3) patient engagement in research funding processes. The barriers were: (1) allocation of research funding, (2) identity conflict, (3) applicability of research evidence, and (4) social stigmatization towards autism.

Conclusions: To promote a more equitable approach to research, Western countries' policymakers, researchers and funders should prioritize patient's needs in research-related activities. Adaptations are needed to implement the suggestions in local context.

Keywords: Autism, patient engagement, children population

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2.2 Introduction

Autism spectrum condition (autism) is a neurodevelopmental disability that affects 1-2% of individuals worldwide (American Psychiatric Association [APA], 2013; Elsabbagh et al., 2012). According to the Diagnostic and Statistical Manual of Mental Disorders (Fifth Version), an autism diagnosis is made with individuals who have challenges in social communication and interaction and presented with restricted, repetitive behaviours (APA, 2013). Examples of autistic symptoms that can also be observed in some autistic individuals are avoiding eye contact, following a strict daily routine and getting anxious or upset by minor changes, and experiencing sensory hyper-sensitivity (APA, 2013; Seltzer et al., 2003). These symptoms can impact autistic individuals throughout their life course. A significant proportion of autistic individuals are diagnosed with co-occurring condition as compared to neurotypical individuals, which further impacts their functional abilities and quality of life (Al-Beltagi, 2021). Some of autism comorbidities include attention deficit hyperactivity disorder (ADHD), sleep disorders, anxiety, learning disabilities, dyspraxia, etc. (Al-Beltagi, 2021).

Power dynamics are the unequally shared capacity that exists by virtue of a party's title or social position. The power relations among research participants, community organizations, researchers and funding committees in academic research can impact the kind of research produced and healthcare decisions made (Baum, 2006; Riley, 2003). Compared to traditional research that is led by researchers, patient-oriented research (POR) allows community members to guide and share research that is done "to", "about" or "for" their population. POR are typically co-designed by community members and researchers that levels power dynamics and encourage shared decision-making. It allows meaningful patient engagement, that is to promote community members' sense of belonging in research through engaging them as research participants or throughout the POR process. This is beneficial in prioritizing patients'

values and equipping the patient population and policymakers to make decisions using applicable research evidence generated by the corresponding patient community.

Marginalized populations are groups and/or communities that are discriminated and excluded in the society. Few opportunities are provided to these disadvantaged populations to engage in making social, political, and economic decisions. The unbalanced power relationships among different social groups contributes to marginalization. Based on the literature, some of the recognized marginalized autistic populations include, but are not limited to, groups disadvantaged due to race, sex, gender, sexual orientation, age, culture, language, and/or immigration status (Seng et al., 2012). Marginalized groups often have high risk of developing poor health outcomes, for instance being diagnosed with chronic diseases (Farrell, 2005). Overlapping marginalized social identities could create complicated and unique experiences of social oppression (Crenshaw, 1991). Consequently, extra attention in how intersectionality impacts marginalized groups is needed. Being diagnosed with autism is an identity that often increases one's chances of experiencing social marginalization (O'Connor, 2020). When an autistic individual carries more than one marginalized identity, they often encounter greater disparities in health status and life expectancy than the privileged individuals who hold more power in society (Bishop-Fitzpatrick & Kind, 2017). As a result, it is a priority to address the marginalized populations in autism research.

A good practice of patient engagement in POR should engage the true representation of the targeted population. To engage community members who carry different identities in research, an inclusive research environment and diverse sampling approaches are needed. In the field of autism research, there are concerns that the processes and strategies that were used to meaningfully engage participants in POR systematically under-represented marginalized populations (Fletcher-Watson et al., 2019; McCoy, 2020; West et al., 2016). For example, there is a lack of autism research engaging black and African families (Shaia et al., 2020). Limited

engagement of marginalized populations results in biased research with inaccurate information about the autistic population groups with different identities.

POR that engages children (age 18 or under) is limited due to complicated methodological and ethical considerations (Schmidt & Paris, 1984). Given these challenges, the caregiver with whom an autistic child spends a lot of their time growing up is often engaged in research studying their child instead. Nonetheless, caregivers can only share their best understanding of the youngsters, the caregivers' feedback in research might not always be true to the autistic children and family's personal thoughts. To conduct inclusive POR, studies should engage autistic children's first voice wherever it is ethically possible (Tesfaye et al., 2019).

The promotion of an equity-based approach in research is important when uncovering knowledge that targets additional marginalized identities. This helps to balance the power among different stakeholders when conducting research on autistic children. By enhancing patient engagement, the autism POR produced can better support the marginalized population groups. A scoping review was conducted to investigate who and how were the routinely marginalized autistic children aged 2 to 18 had any form of engagement in academic research, including being research subjects or being part of the POR process. The objectives of this review were (1) to derive some of the under-represented autistic population groups in research by identifying the marginalized autistic children population groups that were included in research, (2) to assess the facilitators and barriers contributing to patient engagement in autistic children's research, and (3) to suggest considerations for a more equitable approach to autistic children's research. It was hypothesized that the different marginalized identities that autistic children carry makes it more challenging for this population to be engaged in research. A secondary analysis will be made to compare research that engaged autistic children's first voice

versus those engaged the caregivers of autistic children. It was hypothesized that children's first voice was under-represented.

2.3 Methods

A scoping review was conducted based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews (PRISMA-ScR) guideline (Tricco et al., 2018). The review protocol was written a priori and approved by McMaster University Master of Public Health program. The protocol is available for dissemination from the corresponding author upon request. Since this study is a review of published studies and does not engage living human or animal participants, there are no major ethical issues to report.

Autism studies that targeted patient engagement published in the last ten years (January 2011-December 2021) in five electronic research databases (Ovid Medline, Ovid Embase, Web of Science Core Collection, Ovid APA PsycINFO and EBSCOhost CINAHL) were screened. The targeted research population was children aged 2 to 18 diagnosed with autism and/or their parents/caregivers. Included studies were primary studies conducted with a qualitative, quantitative or mixed methods study design. Only English language studies were assessed since this was the working language of our research team. Abelson et al.'s three criteria were used to identify literature on patient engagement initiatives that aimed to meaningfully engage marginalized populations; these criteria were: (1) participants received information about an issue; (2) participants had opportunities to discuss the issue amongst themselves and potentially with decision-makers; (3) there was an explicit process for collecting participants' input (Abelson et al., 2010). Studies that did not describe the patient engagement process, were not targeted for actively engaging patients in research in a healthcare setting and/or are a secondary analysis (for example, systematic review or meta-analysis) were excluded. General exclusion

criteria were carefully designed to prevent the promotion of large selection bias, which could limit the generalizability of this review.

Keywords and relevant indexing were developed in consultation with an experienced McMaster University librarian who had specialized training in scientific literature, and further refined through team discussion. To identify potentially relevant research, the search results from the five bibliographic databases were recorded. The final search strategy of these five databases can be found in Appendix A to E, respectively. The latest search was conducted on February 22, 2022.

To increase consistency in the process of selecting sources of evidence among reviewers, a calibration exercise was done with the two reviewers (AL and AK) prior to beginning screening for this review. 50 publications were randomly selected for the calibration exercise. Both reviewers screened the same 50 publications, and they discussed the results. Clarifications on the inclusion and exclusion criteria were discussed and resolved during the calibration exercise. No key changes were made to the screening criteria after completing the calibration exercise.

A data-charting form was developed by the two reviewers to determine the variables for extraction. The form was tested by the two reviewers before use. Data were first manually extracted by the two reviewers independently from the included studies using Microsoft Excel. The completed data-charting form from the two reviewers was then compared; any discrepancies in the form were flagged and discussed between the two reviewers until a consensus was reached. The data-charting form was updated accordingly in an iterative process. To better compare the included studies that were conducted in a first-person perspective with autistic children versus those that studied parents of autistic children, changes were made to the "participant characteristics" table (Appendix F) to include two separated sections. One section is dedicated to extracting available data on the age and gender of the autistic children,

the other section is to extract the same data but on the parents of the autistic children. The structure of Appendix G and H that summarized the facilitators and barriers contributing to autistic participants' engagement in research were discussed with a third researcher (SG) to consider the different ways to present the data.

2.3.1 Data Items

Data on (1) study characteristics, (2) participant characteristics, (3) strengths and limitations in patient engagement, and (4) the facilitators and barriers contributing to participants' engagement in autism-related research were extracted.

For study characteristics, the publication year, country of where the study was conducted, and study design were abstracted. Specifically, interpretations from the two reviewers were needed when the study did not explicitly report its study design.

Regarding participant characteristics, the types of research participants, sampling method, inclusion criteria, age, gender, and the total number of participants who responded to the included studies were recorded. The strengths and limitations in participant engagement that were explicitly stated and acknowledged in the manuscripts by the authors were also cited. The type of research participants can be any stakeholders, for example, service providers/professionals, researchers, and teachers/school personnel, engaged in autism research. Each type of research participant that was engaged in the included studies was recorded, and the group of research participants that match our inclusion criteria was indicated (bolded) in the data table. Only the participant characteristics data corresponding to the type of research participants that fit our search were extracted. The total number of participants who responded is different from the total number of participants enrolled in the study. Those participants who were lost to follow up or did not respond to the study were not considered as actively engaged in the included research studies; thus, these participants' characteristics were not extracted.

The "Why am I always being researched?" guide developed by Chicago Beyond was used for the identification of facilitators and barriers to participant engagement (Chicago Beyond, 2018). This guide is one of the frameworks that addresses the difficulties in engaging the pediatric population in research (Chicago Beyond, 2018). The guide was produced based on the voices of community leaders and organizations, instead of researchers or funders. It suggests seven inequalities that are in the way to generate accurate knowledge of the targeted populations (Chicago Beyond, 2018). This guide is developed aiming to improve youth equity, specifically promoting opportunities to engage in health research and more equitable access to health services among young people in Chicago, United States (US). "Access", "information", "validity", "ownership", "value", "accountability" and "authorship" are the seven inequalities that are held in place by the power dynamics among community organizations, researchers and funders (Chicago Beyond, 2018). "Access" refers to the challenge for members of the community to access creating knowledge about their communities; "information" inequality prevents the community from engaging in research as they do not have enough information to contribute their wisdom to the research development; the "validity" of information shared by the community is often less prioritized than institutional data; community members contributing to research does not have the "ownership" to data produced in the study; the "value" of research for the community is not always clear to the community members; the community "accounts" for the greatest risks as the researchers or funders do not always take the responsibility when the study design does not work; the "authorship" of community organizations in research are restricted and their voices are muted. By addressing these seven inequalities identified by Chicago Beyond and the community organizations which they work with, it is expected that it will tackle the power differences that impact patient engagement in research development (Chicago Beyond, 2018).

The citations of the facilitators and barriers contributing to participant engagement in autism-related research acknowledged by the study authors in the manuscript were extracted. Based on the interpretations of the two reviewers, the context of the cited quotations was grouped by whether it is a facilitator or barrier to patient engagement in research. Thereafter, these quotations were categorized based on which of the seven inequalities were being targeted, then which one of the stakeholder groups under that specific inequality to which the quote belonged referencing the "Why am I always being researched?" guide (Chicago Beyond, 2018). The criteria of what the community organizations, researchers and funders can do to target the seven inequalities in research activities as listed in the guide were summarized in Table 1. The two reviewers used this table to judge how to organize the citations independently and disagreements were resolved until a consensus was reached.

2.3.2 Synthesis of Results

A cross-sectional analysis was conducted to synthesize the results. The data extracted were presented in tables. To meet the objectives of the scoping review, Appendix F includes extracted data of the participant characteristics of the included studies; this includes the type of research participants, sampling method, inclusion criteria, participant's age, gender, and the total number of participants who responded in table 3a, 3b, 3c and 3d, respectively. Consequently, Appendix F is a summary of the characteristics of the participants that were engaged in the included studies. By comparing the characteristics of the participants that were engaged in the included research studies (Appendix F), the primary objective of this scoping review can be addressed by deriving some of the marginalized autistic groups that were under-represented. Table 3a and 3d compare the research that investigated autism from the first-person perspective of autistic children and those that were conducted from the perspective of their parents. Details of the participants' age and gender and the total number of parents and autistic children participants engaged allow us to achieve the secondary objective of this review.

Table 4 includes the strengths and limitations of patient engagement recognized by the study authors, data are presented by citing the exact quotes from the manuscript of the included studies. This also help to supplement the characteristics of the marginalized autistic populations that were under-represented in autistic children's research. Tables 5a and 5b categorize the patient engagement facilitators and barriers compared to the criteria made by the Chicago Beyond guide on how to tackle the seven inequalities from the perspective of three types of stakeholders (Chicago Beyond, 2018). Based on these identified facilitators and barriers, suggestions on how to conduct equity-based patient engagement in autism children's research were made.

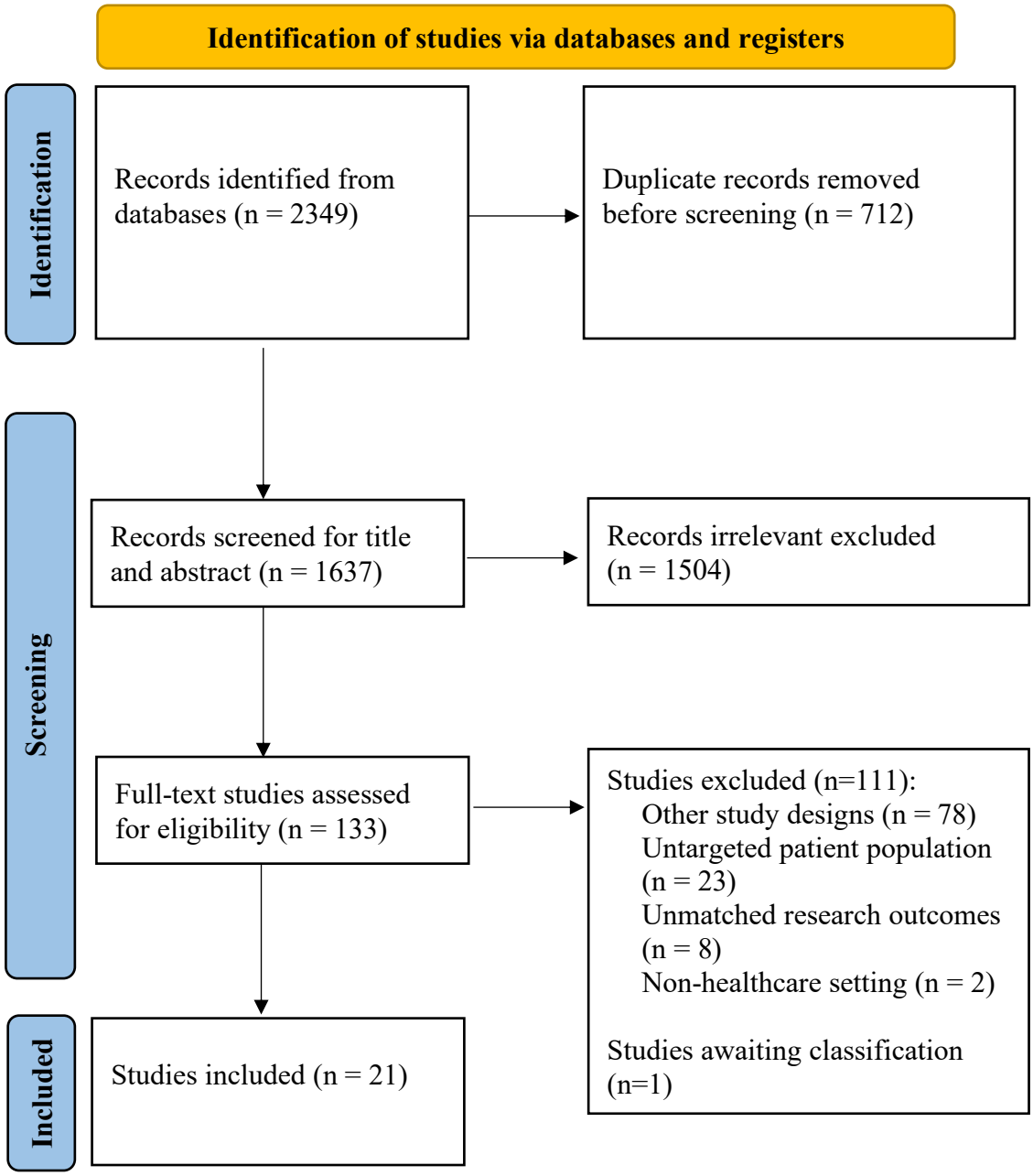
2.4 Results

Screening of sources of evidence followed the PRISMA 2020 flow diagram, which was summarized in Figure 1. A total of 2349 references resulted from the search of the five databases. The final search results were exported into the Covidence system. 712 duplicates were removed. The two reviewers (AL and AK) reviewed and confirmed the duplicates suggested by the Covidence system. The remaining 1637 studies were included for screening. Thereafter, the titles and abstracts of all 1637 studies were screened independently by reviewers AL and AK. Discussions were held to resolve disagreement until a consensus was reached. 1504 studies were excluded after this screening stage. The remaining 133 studies were assessed for full-text eligibility. Similar to the screening of titles and abstracts, both reviewers screened independently, the ambiguities were compared, and a consensus was obtained among the conflicts. After a full-text review, 111 studies were excluded. 78 were excluded as the studies involved secondary analysis (for example, systematic review or meta-analysis) or the study design was not patient-oriented. Most of the studies did not include enough details in their title or abstract to judge whether their patient engagement activities fit our inclusion criteria, so those studies were only excluded after the full-text review were done. 23 studies engaged other

patient populations, either the autistic children participants' age is out of the range, or the parents of an autistic child were not the targeted study participants. Eight studies were excluded because the outcomes were not focused on engagement in research. Two studies were excluded as the research was conducted in a non-healthcare setting, one that does not focus on improving how the autistic symptoms affect the child and their parents. Out of the 133 studies, the publication related to one of the research studies (Olinger, 2011) was not available even through an interlibrary loan request. Therefore, the two reviewers agreed to consider this research as "awaiting classification". This resulted in 21 studies that fit our inclusion criteria.

Figure 1

PRISMA Flow Diagram



Although 21 studies resulted from the search, we adjusted the number of studies included in this review based on the type of data we were extracting. Where applicable, duplicated data were combined and data from different sections of one study was reported separately. Three of the included studies were likely conducted with the same participant samples addressing the Somali community in Bristol, United Kingdom (UK). Ellen Selman et al. and Fox et al. shared the same sampling method (recruitment via Autism Independence's social media network), very similar inclusion criteria and the same age and gender profile of their research participants. The same team of researchers (Nura Aabe, Fiona Fox, Dheeraj Rai, Sabi Redwood and Katrina Turner) conducted more than one of the three studies (Aabe et al., 2019, Ellen Selman et al., 2018 and Fox et al., 2017). Given that the focus of our review was to look at how patient engagement was done in the studies, we treated these three studies as in one setting and referred to them as "Aabe et al. (2019)/ Ellen Selman et al. (2018)/ Fox et al. (2017)" when duplicated data were extracted from these three studies. This was to prevent the effect of these duplicated descriptions of the same group of participants from being exaggerated and skewing our data analysis. On the other hand, Grinker et al.'s research was conducted with two groups of participants in two countries (South Korea and South Africa) (Grinker et al., 2012). Given that the two groups of research participants were different, we presented the data extracted from this study separately from two studies, separated by the section of the study that was conducted in South Korea (SK) and South Africa (SA). When different data were recorded for these two sections, they will be referred to as "Grinker et al. (2012) (SK)" and "Grinker et al. (SA)" respectively. In general, there are 20 studies to consider for study characteristics (Table 2) and participant characteristics (Table 3) data; data regarding the strengths and limitations in participants engagement and facilitators and barriers of patient engagement in research will be extracted from 21 studies.

The characteristics of each of the included studies, including the publication year, the country where the study was conducted and study design, are illustrated in Table 2 Study Characteristics (presented in the alphabetic order by the first author's last name). Whilst our screening criteria included studies that were conducted in the past ten years (2011-2021), most of the studies that met our screening criteria are published in the later half of the targeted ten years. The study with the earliest publication date is 2012. However, only two of the studies were published in 2012, and all of the remaining studies (90.5%) were published after 2015, that is 2016 (n = 1), 2017 (n = 4), 2018 (n = 1), 2019 (n = 2), 2020 (n = 4) and 2021 (n = 7). Regarding the country where the study was conducted, some of the included studies were conducted across multiple countries. Among the included studies conducted in 20 settings (Table 2), most of the studies were conducted in the US (n = 9, 45%) and the UK (n = 5, 25%). Other countries that were occasionally engaged were Canada (n = 3), Australia (n = 2), Spain (n = 2), France (n = 2), Ghana (n = 1), Czech Republic (n = 1), Finland (n = 1), Israel (n = 1), Macedonia (n = 1), Norway (n = 1), Poland (n = 1), Portugal (n = 1), South Korea (n = 1) and South Africa (n = 1). It is important to be aware that this calculation is considering Fletcher-Watson, 2017 for engaging participants from 11 European countries and most of the participants engaged in Pohl et al., 2020 were from five different countries. No quantitative studies were conducted among those studies that fit our inclusion criteria; only qualitative (n = 10) and mixed methods (n = 10) were used.

Participant characteristics were extracted based on the type of research participants engaged (Table 3a), sampling method (Table 3b), inclusion criteria (Table 3c), age and gender of the autistic children and their parents who were actively engaged in the study (Table 3d), and the total number of participants responded in the study (Table 3d). Appendix F showed an overview of the participant characteristics of these 20 settings by combining the data presented in tables 3a, 3b, 3c and 3d.

Table 3a shows that parents/caregivers/families of an autistic child were one type of stakeholder that was commonly investigated in autism patient engagement studies. All 20 settings (100%) engaged parents, caregivers or families of an autistic child, but only four out of the 20 settings (20%) also engaged autistic children (Ami-Narh, 2019; Anderson et al., 2021; Groba et al., 2021; Linder et al., 2021). None solely focused on autistic children.

In Table 3b, out of seven groups of research participants in the 20 settings that explicitly stated their sampling methods, purposive sampling (n = 4) and convenience sampling (n = 3) were often used. Nine groups of research participants included a description of their sampling procedures, yet the exact sampling methods were not mentioned. Based on the reviewers' interpretation, all of these sampling procedures (n = 9) were attempting to describe the nonprobability sampling techniques (purposive sampling or convenience sampling) (Etikan et al., 2015).

In Table 3c, the inclusion criteria revealed the main characteristics of the patients that the studies were targeting for engagement. All patient groups (n = 26, 100%) included those who had personal experiences with autism or with an autism diagnosis in their inclusion criteria. Some of the factors that were commonly listed in the inclusion criteria of the 20 settings include (1) limiting to a specific geographic area/organization/service providers (for example, treatment centre, school/specific education centre) (n = 11, 55%), (2) age range of autistic child (n = 6, 30%), (3) race/ethnicity (n = 3, 15%), (4) specific time of when autism diagnosis was made (n = 3, 15%), (5) language requirement (n = 2, 10%), (6) gender (n = 2, 10%), (7) enrolled in financial support program or had specific household income requirements (n = 1, 5%), and (8) had access to technological devices or show sufficient technological competency to get engaged in research (n = 1, 5%). Only one setting (Linder et al., 2021) had a relatively flexible inclusion criterion directed toward parents and young autistic adults that have personal experiences with autism.

Table 3d captured the total number of participants who responded in the study, age range, mean age and gender by percentage (%) of male autistic child and their parents. The number of participants engaged in five sections of the 20 settings was unknown. However, considering those that were known, the total number of participants included in the 20 settings was at least 2027 participants, which was predominantly contributed by the number of parents engaged in the "survey" sections in Fletcher-Watson, 2017 (n = 1040, 51.3%). The total number of parents (at least n = 1995) engaged in the 27 out of 30 sections of research was contrastingly different from the total number of autistic children engaged (n = 32) in three sections of research. Regarding the age range of all parent participants, the minimum age was 20 and the maximum was 68. The calculated average age of engaged parents based on "age range" and "mean age" was both around 44 years old. The calculated average "% of male parents" engaged was 21.6%, meaning that the research participants were mostly made up of females. The age range of the autistic children who were directly engaged in the study or engaged through their parents' engagement was variable. It ranged from age 2 to 18, which covered the entire targeted age range of our review. The calculated average of "autistic children mean age" was 8.6 years old. Most of the autistic children engaged were male, the derived average "% of male autistic children" was 87.6%. Of those research that shared age and gender information of the parents, some of the research sections (n = 18, 50%) did not provide any information on the age or gender of their autistic child. In general, a few sections of the study (n = 8, 22.2%) did not provide any information on the age and/or gender of their participants (parents or autistic children). Pohl et al., 2020 was the only included study that explicitly collected information on the participants' (mothers of autistic children) sex and gender. Table 3e summarized the details of the calculations and the calculated data.

Table 4 illustrates the strengths and limitations related to participant engagement in the included studies that were acknowledged by the authors. The data extracted were quotations of

the strengths and limitations that were explicitly written by the authors in the manuscript. Efforts have been made to ensure that only the study strengths and limitations listed related to participant engagement were extracted. Overall, there was a lack of patient engagement strengths mentioned by the authors. Only 5 out of 21 included studies (23.8%) mentioned a patient engagement strength. Each of these studies listed one strength. Of these five strengths, the authors engaged participants by (1) addressing the power dynamics among the different participant groups or between the participants and the researchers (n = 2) and (2) were culturally sensitive and responsive to the needs of the local community (n = 3). 18 out of 21 included studies (85.7%) included at least one limitation of their patient engagement process, which contrasted to the few strengths mentioned by the studies. A total of 37 limitations (listed in bullet points in Table 4) were extracted from the studies. All concerns were that the patients engaged in the study were not a good representation of the population (n = 37, 100%), which contributed to the lack of diversity of participants. This led to an underpowered study with potential bias and affects the applicability of the study results. The reasons that were attributed to this concern were (1) the lack of control samples, (2) small autistic sample size, (3) engaging participants of one gender over others, (4) engaging participants of a certain age range, (5) engaging participants only if they meet certain language requirements, and (6) engaging participants of a certain geographical location/country, community/treatment centres or of a specific race/cultural background were targeted. 4 out of 21 included studies (19%) did not include any information on its patient engagement strengths and limitations. There were no suggestions made by the studies on how to improve their patient engagement, except Ogourtsova et al., 2021 that suggested recruiting members with diverse characteristics by broadening the recruitment strategies.

Table 5 categorized the facilitators (Table 5a) and barriers (Table 5b) to patient engagement of the 21 included studies based on the "Why am I always being researched?"

guide, targeting the seven inequalities from the perspective of three stakeholder groups. Appendix G and H included the cited quotations of facilitators and barriers data, respectively, in addition to what Tables 5a and 5b captured. Some of the data engaged both community organizations and researchers, thus these two stakeholders were sometimes combined to better reflect the facilitators/barriers that engaged more than one category of stakeholders for each inequality. 15 facilitators were extracted (Table 5a); eight barriers were extracted (Table 5b). No facilitators that improved inequality "information", "validity", "ownership" and "authorship" were identified at the funders level. No barriers related to "funders" in all seven inequalities were identified; no barriers in all three stakeholder groups were acknowledged by the study authors relating to "ownership" and "authorship"; no barriers corresponding to "information", "validity" and "value" were extracted at the "researchers" level.

2.5 Discussion

2.5.1 Summary of Evidence

This scoping review examined patient engagement of marginalized populations in autistic children's research. Through this review, we aimed (1) to identify the marginalized autistic populations that have been included in autistic children's research so to acknowledge some of the under-represented autistic population groups in research, (2) to assess the facilitators and barriers contributing to patient engagement in autistic children's research, and (3) to suggest considerations that can be addressed for a more equitable approach to autistic children's research. The primary hypothesis is that autistic children who also carry other marginalized identities were more likely to be excluded from research. The secondary analysis compared research that engaged children's first voice versus those included studies that engaged their caregivers, it was hypothesized that children's first voice was under-represented in research.

A total of 21 studies out of 2349 articles from five research databases fit the inclusion criteria of this scoping review. One of the main reasons why there were a few studies that made it to full-text review but were excluded after this stage of the review was that most of the studies did not provide enough details of their patient engagement initiatives for the reviewers to judge in the title and abstract. The lack of patient engagement and its details in research is the most common exclusion reason in data screening. One of the studies was considered as "awaiting classification" because our team cannot access the full text for review even through assistance from the McMaster University Health Sciences Library interlibrary loan.

The study characteristics of the 21 included studies were recorded. There was an increase of relevant studies published in the past five years (2015-2021); this includes 90.5% of the included studies. Most of the studies were conducted in high-income, Western countries, specifically in the US (45%) and the UK (25%). This reveals that under-developed, developing, and non-Western countries were rarely engaged. There were no quantitative studies, and qualitative (n = 10) and mixed methods (n = 10) study designs were both commonly used. All studies used nonprobability sampling techniques, including purposive sampling and convenience sampling (Etikan et al., 2015). Therefore, it suggests that nonprobability sampling techniques were always used in this research topic. Nonprobability sampling techniques are usually used when randomization is impossible because of a very large population and when the researchers have limited resources (Etikan et al., 2015). However, a major concern with this sampling method is that it does not generate a good representative of the population because the subjective nature in choosing samples (Etikan et al., 2015). This means that the use of nonprobability sampling techniques in these included studies raised concerns in biased patient engagement. A total of at least 2027 participants were engaged, this includes at least 1995 (98.4%) parents and 32 autistic children (1.6%). However, most of the participant groups were small scale, each engaged only 3 to 335 participants. The age range of engaged parents

was 20 to 68 years old, and the autistic children engaged were aged 2 to 18. Most of the parents engaged were females (only 21.6% were male). Oppositely, most of the autistic children engaged were males (87.6%). Most of the studies only included the gender of the engaged participants, except Yingling et al., 2020 which collected information on the participant's sex and gender. Considering one's sex and gender could be different, it is important for studies in the field to consider surveying data of participant's sex and gender. The two major patient engagement strengths acknowledged by the authors of the studies were that they addressed power dynamics and the engagement process was culturally sensitive and responsive. This aligns with the values of POR. The limitations to patient engagement were (1) the lack of control samples, (2) small autistic sample size, (3) unbalanced participant engagement of one gender over the others, (4) engaging participants of a specific age range, (5) engaging participants only if they meet certain language requirements, and (6) participants of a certain geographical location/country, community/treatment centres or of a specific race/cultural background were targeted.

Considering the data on study and participant characteristics, there was an under-inclusion of autistic children with marginalized identities in patient engagement, which supports the primary hypothesis. The marginalized autistic populations engaged in the 21 studies were (1) those living in Western countries, (2) those who received autism services from treatment centres and schools/specific education centres that collaborate with researchers, (3) autistic preschoolers/toddlers and adolescents, (4) European/White, Somali, Ghanaian and Korean parents, (5) parents whose child received an early or recent autism diagnosis, (6) bilingual Zulu-/English-speaking parents and those with sufficient English to participate, (7) female parents of autistic children, (8) male autistic children, (9) those enrolled in governmental healthcare financial support program, and (10) those who have access to a computer/smartphone and were comfortable with technological applications. Comparing the

identities of the participants engaged in these studies with the identities that were known to be disadvantaged in our society (race, sex, gender, sexual orientation, age, culture, language, and/or immigration status), we derived some of the under-represented marginalized autistic populations: (1) those living in developing or under-developed countries, (2) those who received autism services from treatment centres and schools/specific education centres that do not have existing connections to collaborate with researchers, (3) families of ethnic minority in Western countries, (4) autistic children who received their diagnosis later in their childhood, (5) families whose first language is not English, (6) male parents of autistic children, (7) female autistic children, (8) families with low household income who are not enrolled in governmental healthcare financial support program and (9) those who do not have access to technological devices or lack technological literacy skills.

To conduct equity-based research, future studies should enhance engagement of these under-represented marginalized autistic populations. It is important to be aware that these are just some of the examples of the many marginalized autistic populations that are under-represented in research, but it does not reflect all the marginalized autistic populations. More research engaging these under-represented autistic populations is required, so that researchers produce accurate knowledge that can support policymakers' decision-making process. Otherwise, the decisions made are limited by the lack of data conducted in this area, which is one of the major limitations of this review. As a result, policymakers and funders should support researchers to engage marginalized populations in their research.

Secondary analysis revealed that there was an under-represented children's first voice engaged in autism research as a research participant or in the POR process, and their feedback was often replaced with engaging their caregivers instead. A total of at least 2027 participants were engaged in the 21 included studies, including at least 1995 (98.4%) parents and 32 autistic children (1.6%). None of the included studies engaged autistic children as the sole research

participant while all of the included studies engaged parents. Being an autistic child is a marginalized identity that makes it more challenging for this population to be engaged in research. Groba et al., 2021 suggested that this is mostly due to the ethical considerations of engaging autistic children, which is in line with our predictions.

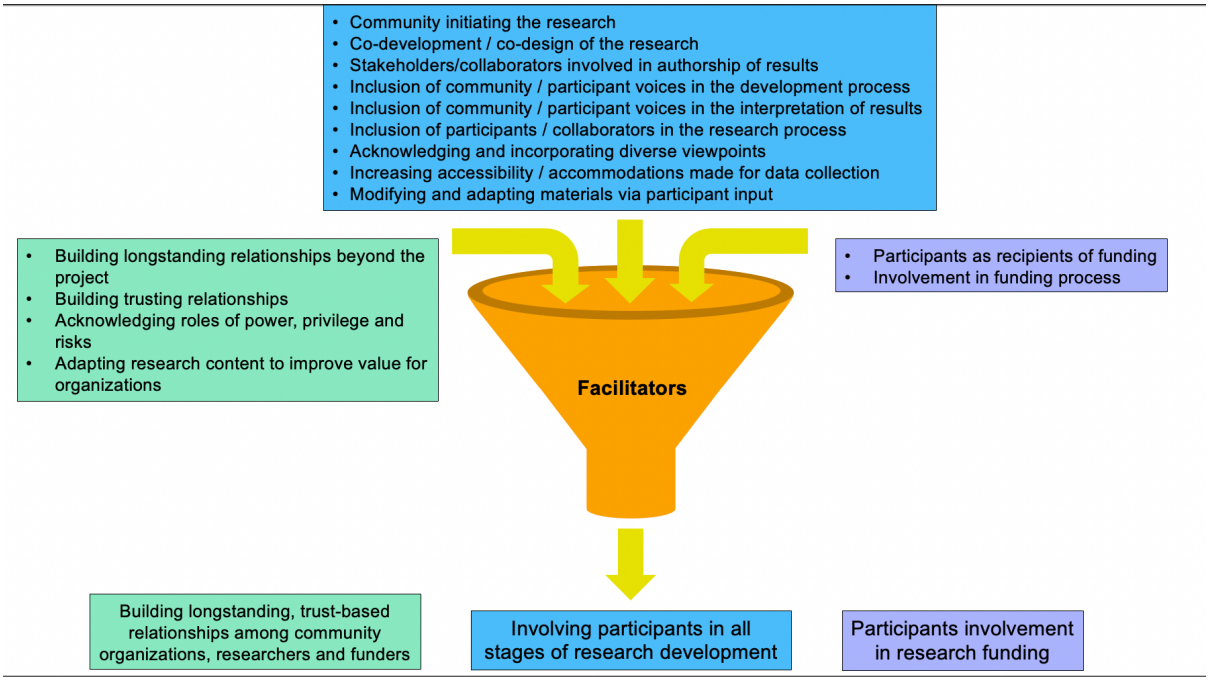
Based on the identified facilitators targeting the seven inequalities at the level of community organizations, researchers and funders, we developed a set of recommendations for health policymakers, researchers and funders to consider when promoting participant engagement in POR. Figure 2 summarizes the facilitators in patient engagement at the top of the funnel diagram, and the derived recommendations for conducting equity-based patient engagement research are at the bottom of the funnel.

Firstly, we suggest actively engaging autistic children in all stages of research development, from the initiation of research, study design, data collection, data interpretation to results dissemination. Considering the different stages of research development is a helpful guide to increasing patient engagement in research. Specifically, of the 21 included studies, there was a lack of patient engagement when identifying research priorities and designing the research. Researchers can reference Aabe et al., 2019/Ellen Selman et al., 2018/Fox et al., 2017 on how to engage members of the community during the initiation phase of the research, specifically in conducting the research based on their priorities. Secondly, building longstanding, trust-based relationships among community organizations, researchers and funders is vital. Acknowledging the roles of power, privilege and risks among all stakeholders is essential for engaging participants in research, and efforts to balance these factors are needed. Researchers shall be patient with developing a research-participant relationship. Recognizing the different influencers and dimensions of trust can aid stakeholders to effectively target ways to enhance trust when connecting with each other in research, even when working with limited time and resources (Wilkins, 2018). Funding activities for the dissemination of research results

can help maximize the use of the data produced in POR. Lastly, we recommend engaging participants in the funding discussion panel to decide how to allocate research funding. The conflicts experienced from the dual roles of being a community member and a co-researcher should also be described under "conflict of interest". James, 2021 and Strange et al., 2020 provided funding incentives to encourage participant engagement, yet studies that provided monetary incentive needs to be cautious of contributing to unfair inclusion of those with limited financial resources.

Figure 2

Overview of Facilitators in Patient Engagement and Derived Recommendations



Note: At the top of the funnel diagram are the facilitators to patient engagement extracted from the engaged studies, and the derived recommendations are at the bottom of the funnel. The recommendations are generated by grouping the facilitators that are of similar themes together. The corresponding recommendations made were based on each of the three groups of facilitators in the same colour, respectively.

Based on the barriers to patient engagement, we developed the following suggestions to policymakers, researchers and funders to improve patient engagement. Figure 3 summarizes the barriers to patient engagement and corresponding recommendations. The same process as facilitators were used to generate these recommendations.

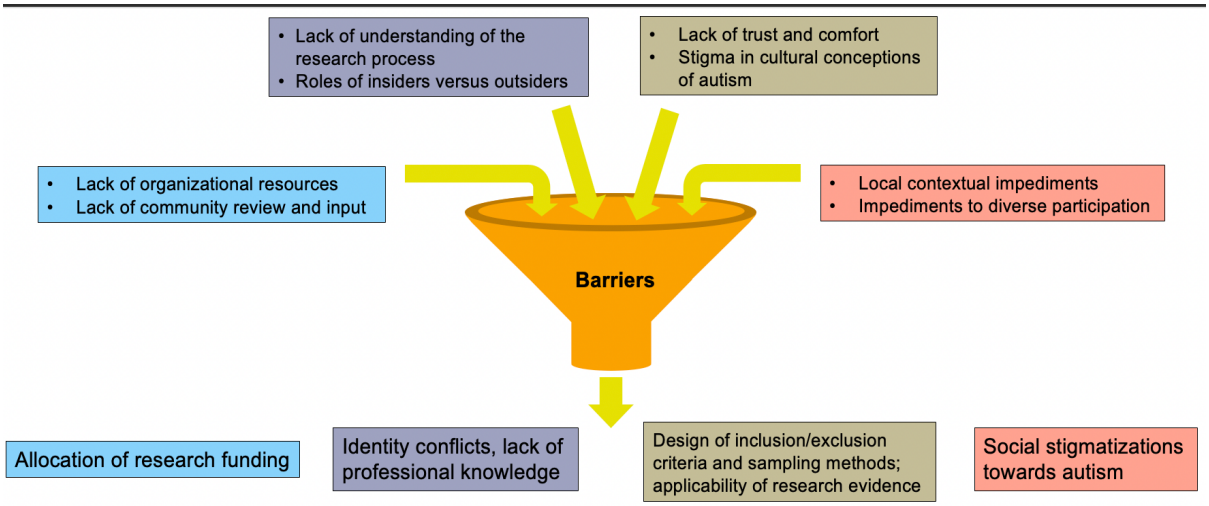
Firstly, the lack of diverse identities of participants engaged was associated with the limited applicability of data produced from the research. This can be improved by reminding the researchers to carefully consider the design of inclusion criteria, exclusion criteria and sampling method when developing research. Researchers should be aware that how they design the inclusion criteria and sampling method directly impacts the participants engaged in the study. They should follow ethical guidance to develop "appropriate inclusion" and avoid designing "inappropriate exclusion". Therefore, inappropriate exclusion of children, including autistic children's first voice, should be avoided. On the other hand, funders should be aware of the specific requirements they are setting up for their research grant applications. Only factors that are essential for the topic of research should be listed as the criteria for the research; this can better reflect the reality and respect the intersected identities that participants carry. When inappropriate inclusion/exclusion criteria and sampling methods are set up by the researchers or funders, this directly causes systematic exclusion of some patient populations from engaging in research. Also, when policymakers make decisions based on research evidence conducted with representative samples, the participants engaged in the research should be carefully reviewed. For example, if the research includes a specific group of autistic children, policymakers should only consider this piece of evidence for decision-making engaging the specific autistic children's population and not generalize the data for making decisions for the entire autistic children's population. When making decisions targeting the general autistic children's population, research evidence generated from studies that used broad inclusion criteria that capture autistic children population that has different marginalized

identities is more reliable. Secondly, there was a lack of resources to support community organizations to share their feedback in research. When applying for research funding, requirements can be set up to ensure that sufficient funding is allocated to community organizations to include their input in the research study, instead of sending all the project funding to researchers directly. This funding method can facilitate collaboration between community organizations and researchers, thus increasing the engagement of patients in research. Thirdly, the identity conflicts of being a member of the community and a researcher who conducted the study is a challenge to participant engagement in research. This could be due to community members' lack of understanding of how research is conducted. Therefore, we suggest that after participants showed interest in research engagement, researchers can start the conversation by sharing the different research tools that are available for use to support the community members to conduct their research. This kind of conversation can also help to explore the participant's priorities and interests, facilitating them to take the lead in initiating the research. Public health officers should also invest in developing educational courses to empower community members to be "expert patients". Lastly, more work needs to be done to improve participants' trust and comfort around engagement in research. The stigmatization of autism in our society is one of the important factors that contributes to this challenge. Tackling the stigmatization of autism should be a priority for our society to increase engagement of autistic individuals in research. Increasing acceptance of autism can be done by educating the public with the most updated information about autism. Policymakers can engage autistic individuals when designing these initiatives as it is important to tailor the content towards the myths that contributed to the discriminative behaviors that autistic individuals experienced. Addressing the specific barriers related to the marginalized identities shall support policymakers to effectively increase engagement from these communities. This, again, reveals the importance of engaging patients in research. Improving the negative image of research in

the society is essential. Providing education to journalists and engaging them in this kind of work can be a helpful first step.

Figure 3

Overview of Barriers in Patient Engagement and Derived Recommendations



Note: At the top of the funnel diagram are the barriers to patient engagement extracted from the included studies, and the derived recommendations are at the bottom of the funnel. The recommendations are generated by grouping the barriers that are of similar themes together. The corresponding recommendations made were based on each of the three groups of facilitators in the same colour, respectively.

Our recommendations are based on limited data addressing the role of funders in patient engagement. Efforts need to be made to encourage reporting of funding information. Not only to declare financial conflicts in the "conflict of interest", "disclosure statement" or "acknowledgement" section of a manuscript, descriptions of how funding guided the research development should also be reported. A clarification of the "funding" section in a manuscript should be made. Additionally, overlapping facilitators/barriers between community organizations and researchers were often recorded in the extracted data. This is reasonable as

it aligns with the value of POR, which is supporting community members to take lead in conducting research that is related to them.

To conclude, improvements need to be made to enhance who and how routinely marginalized autistic children (aged 2 to 18) are engaged in research. An equity-based approach is needed to avoid systematically limiting marginalized autistic children from being engaged as a research participant or in the POR progress. This is vital in producing research evidence that is applicable to autistic populations. Our review identified some of the marginalized autistic populations that future studies should attempt to include and encourages more research that engages children's first voice. We also suggested some considerations derived from the extracted data on how to improve patient engagement in autistic children's research. Collaboration among policymakers, community organizations, researchers and funders is needed to conduct inclusive autistic children's research.

It is important to be aware that the seven recommendations do not work independently, rather they work collaboratively in improving patient engagement in POR. For instance, solely implementing appropriate inclusion criteria and avoiding inappropriate exclusion criteria that facilitates the engagement of a more equitable representative population in research is not enough to improve patient engagement in POR as it lacks the elements to actively engage the community members in research. On the other hand, engaging participants in all stages of research development, building longstanding, trust-based relationships among the stakeholders and empowering patient to guide their research can improve patient's sense of belonging in the research project upon they are equitably included in the study. Oppositely, if the inclusion criteria, exclusion criteria and sampling method of POR were not probably designed, it results in systematically biased sample of the population even other recommendations are implemented. As a result, when all seven recommendations are implemented, it is expected that they will create a synergy effect to promote patient engagement in autistic children's

research. Moreover, the marginalized identities, facilitators and barriers to patient engagement in POR, and derived recommendations resulted from this review shall also be applicable to POR that study other health conditions, especially in paediatric health research. Consequently, the result of this review has potential implications in changing how research is conducted and healthcare decisions are made.

2.5.2 Limitations

Despite efforts made by our team to minimize selection bias and enhance the generalizability of this review, the inclusion and exclusion criteria we set up still limited our search results. There were many different types of stakeholders, for example, providers/professionals, researchers, and teachers/school personnel, that were engaged in autistic children's POR. However, our research only looked at autistic children and their parents/caregivers. Consequently, the data extracted are only related to these two groups of participants. Since our recommendations for enhancing patient engagement in research are developed based on these data, a limitation of the recommendations made here is that it is only applicable to these two groups of participants. Future studies can consider reviewing other stakeholders engaged in autistic children's research. Also, we set an age range for the autistic children that we targeted in this review, it was aged 2 to 18. We made this decision based on the evidence that autism is diagnosed as early as age two and initial search from further limiting the age range reveals that there is very limited patient engagement research done with specific age range of pediatric autism populations. Consequently, the lack of information increases challenges and risks of engaging young autistic children's first voice, especially on how to support young autistic children, non-verbal autistic individuals and autistics individuals with high support needs to engage in research. It is more reasonable for future research to start with focusing on engaging autistic youth's (aged 13-18) first voice in research upon more autism

research are conducted with this group. When more evidence is available to judge the benefits and risks of engaging young autistic children (aged 12 and under) in research, it will be beneficial to re-investigate the importance of engaging this group in research and how can this be done efficiently and ethically. We are also aware that some research uses different age ranges for the pediatric population, for example, "age 16 or below" or "up till age 24". Whilst our inclusion criteria can also cover those studies that set the age limit up to age 16, we were limited from covering autistic young adults aged 19-24. Additionally, since the working language of our team is English, we limited our review to studies that were written in English. This could be the reason why there was a lack of data from under-developed and developing countries, and most of the data are from high-income, Western countries where researchers are proficient in English. If there are sufficient resources, translators can be invited to join the team. This will allow broadening of the language requirements and the inclusion of studies that are written in other languages.

Olinger, 2011 was categorized as "awaiting classification" since our team cannot access the full-text manuscript for screening despite a request through interlibrary loan. Based on the information available in the abstract, there is a high chance that this study will be included in our review if we have access to the full text for review. If the study were included in the review, its data on study characteristics and participant characteristics would have aligned with the data trend collected from the 21 included studies. Information on the strengths and limitations in patient engagement and facilitators and barriers of patient engagement in research are not available via its abstract. This study would have added nine parents/caregivers to the total number of parents engaged, and there were no signs that any autistic children were engaged (Olinger, 2011). This would have aligned with the secondary hypothesis.

The recommendations we made are only applicable to Western, high-income countries, and are not likely to be applicable internationally. The data that we collected were studies that

were conducted mostly in high-income Western countries, and this is what we based on to derive the recommendations for patient engagement in autistic children's POR. The lack of data collected from other countries, especially under-developed and developing countries, makes our data not applicable to these geographical locations. Although our data can mostly be applicable to only in Western countries, we did not modify our research questions by limiting to only reviewing data from high-income Western countries because we believe that it is also important to show the kind of research that is lacking in this field. The recommendations should also be used with caution since modifications might be needed. For instance, each country has a different health system, and the recommendations made here provided a general direction of what policymakers, researchers and funders could do to improve patient engagement in local research. An effort has been made to ensure that it is not too specific, so policymakers should reference different implementation frameworks and theories to facilitate the implementation of these recommendations in their local context.

As discussed, there was a lack of facilitators and barriers data corresponding to funders. This also suggests that the recommendations we made are not as applicable to funders. However, according to the "Why am I always being researched?" guide, community organizations, researchers and funders work together to play an important role in patient engagement research (Chicago Beyond, 2018). Therefore, we believe that the recommendations we made are also important for funders to be aware of.

Lastly, it is expected that there will be challenges to implement the derived recommendations. Some of the equity issues, for instance the long history of only engaging male participants in research, is deeply rooted in the society and the research field, thus it will not be easy for our current system to make this change (Holdcroft, 2007). Incorporating these recommendations in healthcare guidelines can facilitate the translation of these recommendations into actions, so to promote patient engagement in research in clinical and

public health practices. Also, there is a lack of assessment on the effectiveness of conducting POR, specifically none of the 21 included studies assessed whether patient engagement benefited the patient, for example improving patient's health outcomes, thus there is a need to conduct this kind of research, both quantitatively and qualitatively. This type of research evidence can support the importance of implementing patient engagement in research and facilitate this systemic change to take place. It should be expected that this kind of systemic changes will not happen in a short period of time, yet it is important to start making the changes now to ensure that current and future research prioritizes patient's needs. The recommendations produced in this review serve as some of the guidance to start initiating this change.

2.6 Funding

No research funding was required to conduct this review. The two reviewers and the entire research team used their personal laptops bought with their own expenses to complete the work for this research project. The Covidence system used for study screening was free of charge through McMaster University. Zoom meetings, which are accessible with the McMaster University work account, were conducted for team discussions. If we were to have additional funding for this review, we would have paid for the right of access to the full text of Olinger 2011 for review. However, as discussed, it is expected that including this study is not going to dramatically change the direction of the results of this review.

2.7 References

Aabe, N. O., Fox, F., Rai, D., & Redwood, S. (2019). Inside, outside and in-between: The process and impact of co-producing knowledge about autism in a UK Somali community. *Health Expectations*, *22*(4), 752–760. https://doi.org/10.1111/hex.12939

Abelson, J., Montesanti, S., Li, K., Gauvin, F. P., & Martin, E. (2010). *Effective strategies for interactive public engagement in the development of healthcare policies and programs* (p. 1-52). Ottawa: Canadian Health Services Research Foundation.

Al-Beltagi, M. (2021). Autism medical comorbidities. *World Journal of Clinical Pediatrics*, *10*(3), 15–28. https://doi.org/10.5409/wjcp.v10.i3.15

American Psychiatric Association. (2013). *DSM-V: Diagnostic and Statistical Manual of Mental Disorders*. http://archive.org/details/diagnosticstatis0005unse

Ami-Narh, D. D. (2019). *Cultural Adaptation of the Transitioning Together® Program for Caregivers of Adolescents with Autism in Accra, Ghana*. Michigan State University. Rehabilitation Counselor Education.

Anderson, C. M., Iovannone, R., Smith, T., Levato, L., Martin, R., Cavanaugh, B., Hochheimer, S., Wang, H., & Iadarola, S. (2021). Thinking Small to Think Big: Modular Approach for Autism Programming in Schools (MAAPS). *Journal of Autism and Developmental Disorders*, *51*(1), 193–211. https://doi.org/10.1007/s10803-020-04532-1

Baum, F., MacDougall, C., & Smith, D. (2006). Participatory action research. *Journal of Epidemiology and Community Health*, *60*(10), 854–857. https://doi.org/10.1136/jech.2004.028662

Bishop-Fitzpatrick, L., & Kind, A. J. H. (2017). A Scoping Review of Health Disparities in Autism Spectrum Disorder. *Journal of Autism and*

Developmental Disorders, *47*(11), 3380–3391. https://doi.org/10.1007/s10803-017-3251-9

Brookman-Frazee, L., Stahmer, A. C., Lewis, K., Feder, J. D., & Reed, S. (2012). Building a research-community collaborative to improve community care for infants and toddlers at-risk for autism spectrum disorders: A Research-Community Collaborative for ASD. *Journal of Community Psychology*, *40*(6), 715–734. https://doi.org/10.1002/jcop.21501

Chicago Beyond. (2018). *Why am I always being researched?* (p. 111). https://chicagobeyond.org/wp-content/uploads/2019/05/ChicagoBeyond_2019Guidebook.pdf

Crenshaw, K. (1991). Mapping the Margins: Intersectionality, Identity Politics, and Violence against Women of Color. *Stanford Law Review*, *43*(6), 1241–1299. https://doi.org/10.2307/1229039

Ellen Selman, L., Fox, F., Aabe, N., Turner, K., Rai, D., & Redwood, S. (2018). 'You are labelled by your children's disability' – A community-based, participatory study of stigma among Somali parents of children with autism living in the United Kingdom. *Ethnicity & Health*, *23*(7), 781–796. https://doi.org/10.1080/13557858.2017.1294663

Elsabbagh, M., Divan, G., Koh, Y.-J., Kim, Y. S., Kauchali, S., Marcín, C., Montiel-Nava, C., Patel, V., Paula, C. S., Wang, C., Yasamy, M. T., & Fombonne, E. (2012). Global Prevalence of Autism and Other Pervasive Developmental Disorders. *Autism Research*, *5*(3), 160–179. https://doi.org/10.1002/aur.239

Etikan, I., Musa, S. A., & Alkassim, R. S. (2015). Comparison of Convenience Sampling and Purposive Sampling. *American Journal of Theoretical and Applied Statistics*, *5*(1), 1. https://doi.org/10.11648/j.ajtas.20160501.11

Farrell, Ann (Ed.) (2005) *Ethical Research with Children.* Open University Press/ McGraw-Hill Education, UK, Buckinghamshire, Milton Keynes.

Fletcher-Watson, S., Apicella, F., Auyeung, B., Beranova, S., Bonnet-Brilhault, F., Canal-Bedia, R., Charman, T., Chericoni, N., Conceição, I. C., Davies, K., Farroni, T., Gomot, M., Jones, E., Kaale, A., Kapica, K., Kawa, R., Kylliäinen, A., Larsen, K., Lefort-Besnard, J., … Yirmiya, N. (2017). Attitudes of the autism community to early autism research. *Autism*, *21*(1), 61–74. https://doi.org/10.1177/1362361315626577

Fletcher-Watson, S., Adams, J., Brook, K., Charman, T., Crane, L., Cusack, J., Leekam, S., Milton, D., Parr, J. R., & Pellicano, E. (2019). Making the future together: Shaping autism research through meaningful participation. *Autism*, *23*(4), 943–953. https://doi.org/10.1177/1362361318786721

Fong, V. C., Lee, B. S., & Iarocci, G. (2022). A community-engaged approach to examining barriers and facilitators to accessing autism services in Korean immigrant families. *Autism*, *26*(2), 525–537. https://doi.org/10.1177/13623613211034067

Fox, F., Aabe, N., Turner, K., Redwood, S., & Rai, D. (2017). "It was like walking without knowing where I was going": A Qualitative Study of Autism in a UK Somali Migrant Community. *Journal of Autism and Developmental Disorders*, *47*(2), 305–315. https://doi.org/10.1007/s10803-016-2952-9

Galpin, J., Barratt, P., Ashcroft, E., Greathead, S., Kenny, L., & Pellicano, E. (2018). "The dots just don't join up": Understanding the support needs of families of children on the autism spectrum. *Autism: The International Journal of Research and Practice*, *22*(5), 571–584. https://doi.org/10.1177/1362361316687989

Grinker, R. R., Chambers, N., Njongwe, N., Lagman, A. E., Guthrie, W., Stronach, S., Richard, B. O., Kauchali, S., Killian, B., Chhagan, M., Yucel, F., Kudumu, M., Barker-Cummings, C., Grether, J., & Wetherby, A. M. (2012). "Communities" in Community Engagement: Lessons Learned From Autism Research in South Korea

and South Africa: "Communities" in community engagement. *Autism Research*, *5*(3), 201–210. https://doi.org/10.1002/aur.1229

Groba, B., Nieto-Riveiro, L., Canosa, N., Concheiro-Moscoso, P., Miranda-Duro, M. del C., & Pereira, J. (2021). Stakeholder Perspectives to Support Graphical User Interface Design for Children with Autism Spectrum Disorder: A Qualitative Study. *International Journal of Environmental Research and Public Health*, *18*(9), 4631. https://doi.org/10.3390/ijerph18094631

Hallett, V., Mueller, J., Breese, L., Hollett, M., Beresford, B., Irvine, A., Pickles, A., Slonims, V., Scott, S., Charman, T., & Simonoff, E. (2021). Introducing 'Predictive Parenting': A Feasibility Study of a New Group Parenting Intervention Targeting Emotional and Behavioral Difficulties in Children with Autism Spectrum Disorder. *Journal of Autism and Developmental Disorders*, *51*(1), 323–333. https://doi.org/10.1007/s10803-020-04442-2

Holdcroft, A. (2007). Gender bias in research: How does it affect evidence based medicine? *Journal of the Royal Society of Medicine*, *100*(1), 2–3.

James, K. P. (2020). *Cultural adaptations of evidence based practices in supporting children diagnosed with autism spectrum disorder* [Thesis]. https://scholarworks.alaska.edu/handle/11122/12303

Linder, D. E., Folta, S. C., Must, A., Mulé, C. M., Cash, S. B., Halbreich, E. D., Colón, C., Sullivan, S., Sanabria, E., Gibbs, D., & Farrell, T. (2021). A Stakeholder-Engaged Approach to Development of an Animal-Assisted Intervention for Obesity Prevention Among Youth With Autism Spectrum Disorder and Their Pet Dogs. *Frontiers in Veterinary Science*, *8*. https://www.frontiersin.org/article/10.3389/fvets.2021.735432

McCoy, M. S., Liu, E. Y., Lutz, A. S. F., & Sisti, D. (2020). Ethical Advocacy Across the Autism Spectrum: Beyond Partial Representation. *The American Journal*

of Bioethics: AJOB, *20*(4), 13–24. https://doi.org/10.1080/15265161.2020.1730482

O'Connor, C., Burke, J., & Rooney, B. (2020). Diagnostic Disclosure and Social Marginalisation of Adults with ASD: Is There a Relationship and What Mediates It? *Journal of Autism and Developmental Disorders*, *50*(9), 3367–3379. https://doi.org/10.1007/s10803-019-04239-y

Ogourtsova, T., O'Donnell, M. E., Filliter, J. H., Wittmeier, K., BRIGHT Coaching Group, & Majnemer, A. (2021). Patient engagement in an online coaching intervention for parents of children with suspected developmental delays. *Developmental Medicine and Child Neurology*, *63*(6), 668–674. https://doi.org/10.1111/dmcn.14810

Olinger, C. (2010). *Working toward agency among the autism community: A participatory action research study.*

Pickard, K. E., Kilgore, A. N., & Ingersoll, B. R. (2016). Using Community Partnerships to Better Understand the Barriers to Using an Evidence-Based, Parent-Mediated Intervention for Autism Spectrum Disorder in a Medicaid System. *American Journal of Community Psychology*, *57*(3–4), 391–403. https://doi.org/10.1002/ajcp.12050

Pohl, A. L., Crockford, S. K., Blakemore, M., Allison, C., & Baron-Cohen, S. (2020). A comparative study of autistic and non-autistic women's experience of motherhood. *Molecular Autism*, *11*(1), 3. https://doi.org/10.1186/s13229-019-0304-2

Rabba, A. S., Dissanayake, C., & Barbaro, J. (2020). Development of a Web-Based Resource for Parents of Young Children Newly Diagnosed With Autism: Participatory Research Design. *JMIR Pediatrics and Parenting*, *3*(2), e15786. https://doi.org/10.2196/15786

Riley, S., Schouten, W., & Cahill, S. (2003). Exploring the Dynamics of Subjectivity and Power Between Researcher and Researched. *Forum Qualitative Sozialforschung / Forum: Qualitative Social Research*, *4*(2), Article 2. https://doi.org/10.17169/fqs-4.2.713

Schmidt, C. R., & Paris, S. G. (1984). The development of verbal communicative skills in children. *Advances in Child Development and Behavior*, *18*, 1–47. https://doi.org/10.1016/s0065-2407(08)60368-7

Seltzer, M. M., Krauss, M. W., Shattuck, P. T., Orsmond, G., Swe, A., & Lord, C. (2003). The Symptoms of Autism Spectrum Disorders in Adolescence and Adulthood. *Journal of Autism and Developmental Disorders*, *33*(6), 565–581. https://doi.org/10.1023/B:JADD.0000005995.02453.0b

Seng, J. S., Lopez, W. D., Sperlich, M., Hamama, L., & Meldrum, C. D. R. (2012). Marginalized identities, discrimination burden, and mental health: Empirical exploration of an interpersonal-level approach to modeling intersectionality. *Social Science & Medicine (1982)*, *75*(12), 2437–2445. https://doi.org/10.1016/j.socscimed.2012.09.023

Shaia, W. E., Nichols, H. M., Dababnah, S., Campion, K., & Garbarino, N. (2020). Brief Report: Participation of Black and African-American Families in Autism Research. Journal of Autism and Developmental Disorders, 50(5), 1841–1846. https://doi.org/10.1007/s10803-019-03926-0

Smith, T., Iadarola, S., Mandell, D. S., Harwood, R., & Kasari, C. (2017). Community-Partnered Research With Urban School Districts That Serve Children With Autism Spectrum Disorder. *Academic Pediatrics*, *17*(6), 614–619. https://doi.org/10.1016/j.acap.2017.04.017

Strang, J. F., Knauss, M., van der Miesen, A., McGuire, J. K., Kenworthy, L., Caplan, R., Freeman, A., Sadikova, E., Zaks, Z., Pervez, N., Balleur, A., Rowlands, D. W., Sibarium, E., Willing, L., McCool, M. A., Ehrbar, R. D., Wyss, S. E., Wimms, H., Tobing, J., … Anthony, L. G. (2021). A Clinical Program for Transgender and Gender-Diverse Neurodiverse/Autistic Adolescents Developed through Community-Based

Participatory Design. *Journal of Clinical Child & Adolescent Psychology*, *50*(6), 730–745. https://doi.org/10.1080/15374416.2020.1731817

Tesfaye, R., Courchesne, V., Yusuf, A., Savion-Lemieux, T., Singh, I., Shikako-Thomas, K., Mirenda, P., Waddell, C., Smith, I. M., Nicholas, D., Szatmari, P., Bennett, T., Duku, E., Georgiades, S., Kerns, C., Vaillancourt, T., Zaidman-Zait, A., Zwaigenbaum, L., & Elsabbagh, M. (2019). Assuming ability of youth with autism: Synthesis of methods capturing the first-person perspectives of children and youth with disabilities. *Autism: The International Journal of Research and Practice*, *23*(8), 1882–1896. https://doi.org/10.1177/1362361319831487

Tricco, A. C., Lillie, E., Zarin, W., O'Brien, K. K., Colquhoun, H., Levac, D., Moher, D., Peters, M. D. J., Horsley, T., Weeks, L., Hempel, S., Akl, E. A., Chang, C., McGowan, J., Stewart, L., Hartling, L., Aldcroft, A., Wilson, M. G., Garritty, C., … Straus, S. E. (2018). PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. *Annals of Internal Medicine*, *169*(7), 467–473. https://doi.org/10.7326/M18-0850

West, E. A., Travers, J. C., Kemper, T. D., Liberty, L. M., Cote, D. L., McCollow, M. M., & Stansberry Brusnahan, L. L. (2016). Racial and Ethnic Diversity of Participants in Research Supporting Evidence-Based Practices for Learners With Autism Spectrum Disorder. *The Journal of Special Education*, *50*(3), 151–163. https://doi.org/10.1177/0022466916632495

Wilkins, C. H. (2018). Effective Engagement Requires Trust and Being Trustworthy. *Medical Care*, *56*(10 Suppl 1), S6. https://doi.org/10.1097/MLR.0000000000000953

Yingling, M. E., Hock, R. M., Feinberg, M. E., & Holbert, A. A. (2020). Community-

engaged process to adapt evidence-based programs for parents of children with autism spectrum disorder. *Children and Youth Services Review*, *112*, 104876. https://doi.org/10.1016/j.childyouth.2020.104876

2.8 Tables

Table 1

"Why am I always being researched?" guide by Chicago Beyond

Inequalities	Stakeholders	Suggestions
Access	Community organizations	• Recognize that the power dynamic makes it tempting to compromise what matters for the chance to produce research evidence. • Where possible, speak up to participate—or not participate— in research on your own terms, and shape research to help your community.
	Researchers	• Design research to serve community purpose. • Not participate in research that perpetuates the researcher as "brains" and community as "brawn" stereotype. • Insist that conversations about community happen with community.
	Funders	• Fund research that community organizations want, need, and are able to lead. Fund research that informs action on root causes. • Not fund research where the questions asked and the approach hold power dynamics in place. • Insist that conversations about community happen with community.
Information	Community organizations	• Get informed. Know your options, know your rights, know the risks. • Seek and use information to ask questions about methods and inputs.
	Researchers	• Share information, recognizing that without it, the community organization cannot actually consent to the research. • Have reciprocal exchange about methods and inputs.
	Funders	• Ensure accountability for the community organization to understand the options and the risks.
Validity	Community organizations	• Value the validity of your own voices at the table, especially on the questions, the inputs to answer the questions, and how participants experience the research. • Build relationship with the researcher. Check partial truths.
	Researchers	• Recognize how the research frameworks, process and inputs reinforce power dynamics, and bring your creativity to making change. • Build relationship with the community organization. • Check partial truths.
	Funders	• Be accountable for what questions the research you fund asks, and what processes and inputs it is (and is not) validating. • Create accountability for authentic engagement between community and researchers. This will check partial truths.

Ownership	Community organizations	• Recognize that the power dynamic makes it tempting to cede ownership. • Build your ownership of your study, which starts with knowing what you want to learn, and why. It is likely that your organization has the most at stake.
	Researchers	• Invite co-ownership of research, in your processes and legal agreements.
	Funders	• Set expectations for co-ownership of research, in processes and legal agreements.
Value	Community organizations	• Get aware of the potential costs - including intangible costs to your participants, organization and community - and advocate for a full accounting. • Get clear and speak up on how research can produce value for your community.
	Researchers	• Recognize the full cost of the research - including intangible costs to participants, community organization, and community - and find ways to support it. • Shape research so producing value for the community is central.
	Funders	• Account for the full cost of the research—including costs to recruit additional participants, intangible costs borne by participants, staffing at the community organization—and support these costs. • Insist on clarity: How will the research benefit the community? According to whom?
Accountability	Community organizations	• Build trust-based relationships with the other entities. Hold researchers with whom you have built trust accountable. • Identify and mitigate risks to you and to your stakeholders.
	Researchers	• Build trust-based relationships with the other entities. Be accountable to understand the context. • Own your role in missteps. • Help identify and mitigate risks.
	Funders	• Build trust-based relationships with the other entities. Be flexible in timelines so trust can develop. Be accountable to understand the context. • Own your role in missteps. • Help identify and mitigate risks.
Authorship	Community organizations	• Recognize that the power dynamic makes it tempting to cede interpretation and presentation of the results. • Participate in how results are made into meaning, and shared. Is it contextualized? Can you hear your participants?
	Researchers	• Invite co-ownership in contextualizing and sharing results. Analyze and frame data with an equity lens for greater impact.
	Funders	• Set expectations for co-ownership of contextualizing and sharing results. Create accountability for an equity lens for greater impact.

Table 2

Study Characteristics

	Author (y)	**Country**	**Study Design**
1	Aabe et al. (2019)/ Ellen Selman et al. (2018)/ Fox et al. (2017)	United Kingdom (Bristol)	Qualitative study (community-based, participatory research, co-production)
2	Ami-Narh (2019)	Ghana (Accra)	Qualitative study (participatory action research framework)
3	Anderson et al. (2021)	United States (Florida, Massachusetts, and New York)	Mixed method study
4	Brookman-Frazee (2012)	United States (California)	Mixed method study (community-based participatory research)
5	Fletcher-Watson (2017)	11 European countries: Czech Republic, Finland, France, Israel, Italy, Macedonia, Norway, Poland, Portugal, Spain, and the United Kingdom	Mixed method study
6	Fong et al. (2021)	Canada (British Columbia)	Qualitative study (community-engaged approach)
7	Galpin et al. (2017)	United Kingdom (London)	Mixed method study
8	Grinker et al. (2012) (SK)	South Korea	Mixed method study (community-engaged approach)
9	Grinker et al. (2012) (SA)	South Africa	Qualitative study (community-engaged approach)
10	Groba et al. (2021)	Spain	A qualitative, longitudinal, multicentre study (phenome logical approach, user-centred design)
11	Hallett et al. (2021)	United Kingdom (London)	Mixed method, feasibility study (co-creation)
12	James (2021)	United States (American Samoa)	Qualitative study (community-based formative, participatory action research)
13	Linder et al. (2021)	United States	Qualitative, descriptive study
14	Ogourtsova et al. (2021)	Canada (4 provinces: British Columbia, Manitoba, Quebec, Nova Scotia)	Cross-sectional, mixed-methods approach
15	Pickard et al. (2016)	United States (Michigan)	Qualitative (research-community partnerships and focus group methodology)

16	Pohl et al. (2020)	From Western countries (top 5 countries: UK, US, Australia, France, Canada)[#1]	Mixed methods (co-developed, community-based participatory research)
17	Rabba et al. (2020)	Australia	Qualitative study (participatory action research, co-design)
18	Smith et al. (2017)	United States (California, New York, Pennsylvania)	Qualitative study (community-partnered participatory research)
19	Strang et al. (2020)	United States (Washington, DC)	Mixed methods (community-based participatory design, co-creation)
20	Yingling et al. (2020)	United States	Mixed methods (co-developed, community-based participatory research)

#1: Pohl et al., 2020 was conducted across 17 countries. Most of the samples were from the top five countries, that is United Kingdom, United States, Australia, France and Canada. Since 89.5% (autistic mothers of autistic child) and 94.5% (non-autistic mothers of autistic child) of study participants were from these five western countries, so these five countries were considered as where Pohl et al., 2020 was conducted for data extraction. Other countries Pohl et al., 2020 engaged were Sweden (n = 3), New Zealand (n = 3), Israel (n = 1), Greece (n = 2), South Africa (n = 1), Serbia (n = 1), Belgium (n = 2), Italy (n = 1), UAE (n = 1), Denmark (n = 5), Costa Rica (n = 1), Brazil (n = 1), Monaco (n = 1), Mexico (n = 2), Ireland (n = 5), Netherlands (n = 4), and Germany (n = 3).

Table 3a

Participant Characteristics (Type of Participants)

	Author (y)	Types of research participants[#1]
1	**Aabe et al. (2019)/ Ellen Selman et al. (2018)/ Fox et al. (2017)**	Somali parents
2	**Ami-Narh (2019)**	**Ghanian families** and their **adolescents with autism**
3	**Anderson et al. (2021)**	Experts: researchers and practitioners
		Focus groups: school administrators, educators, and **parents/guardians**
		Randomized controlled trial: **autistic students**, teachers
4	**Brookman-Frazee (2012)**	BRIDGE collaborative members: community providers, funding agency reps, **parents**, primary professionals, service providers
		Focus group: community providers and **parents of children with ASD**
5	**Fletcher-Watson et al. (2017)**	Focus group: **parents of children with autism**, autistic adults and practitioners from healthcare, education and social support settings
		Piloting survey: autistic adults, practitioners and **parents**
		Survey: practitioners, **parents**, teachers, autistic adults, others/missing
6	**Fong et al. (2021)**	Korean parents of children with ASD
7	**Galpin et al. (2017)**	Parents (biological parent, adoptive parent, foster parents and grandparents) of autistic children
8	**Grinker et al. (2012) (SK)**	South Korea focus group: **parents of children with ASD**, teachers from regular and special education schools
9	**Grinker et al. (2012) (SA)**	South Africa focus group: **parents**, teachers, healthcare professionals, traditional healers, and clergy
		South Africa interviews: bilingual Zulu-/English-speaking **parents**
10	**Groba et al. (2021)**	Professionals with experience in the intervention with people with ASD, professionals with experience in the development and design of technology for people with disability, **family members of people with ASD**, **children with ASD**
11	**Hallett et al. (2021)**	Intervention: **parents of verbal and minimally-verbal autistic children**
		Public and Patient Involvement (PPI) Panels: **parents of autistic children**, autistic adults
		Feasibility study: **parent/carer of an autistic child**
		Semi-structured telephone interviews: **parent/carer of an autistic chi**ld
12	**James (2021)**	American Samoan teachers, **parents** and community members

13	**Linder et al. (2021)**	Service providers, **parents**, **young autistic adult**
14	**Ogourtsova et al. (2021)**	Parent advisory group (**parents/caregivers** of children with disabilities), BRIGHT research team member
15	**Pickard et al. (2016)**	**Parents of a child with a current ASD diagnosis**; professionals who provided direct and parent-mediated interventions to families of children with ASD
16	**Pohl et al. (2020)**	**Autistic mothers** who had at least one autistic child of any age[#2]
		Non-autistic mothers (without a diagnosis of autism) who had at least one autistic child of any age[#2]
17	**Rabba et al. (2020)**	**Parents of young children newly diagnosed with ASD**, health professionals and researchers
18	**Smith et al. (2017)**	School personnel, **parents**, and researchers
19	**Strang et al. (2020)**	Co-occurrence of autism/neurodiversity and gender-diversity (A/ND-GD) in youth and their **parents**
20	**Yingling et al. (2020)**	**Parents**, professionals, researchers

#1: Data of all types of research participants that each of the study engaged were recorded. Of those, the type of participants that aligns with the inclusion criteria of this review (autistic children and their parents) was bolded.
#2: Non-autistic mothers refers to those mothers who did not have a formal autism diagnosis but had at least one autistic child of any age and autistic mothers are those mothers who were diagnosed with autism and had at least one autistic child of any age.

Table 3b

Participant Characteristics (Sampling Method)

	Author (y)	Sampling method
1	**Aabe et al. (2019)**	Somali parents: N/A[#1]
	Ellen Selman et al. (2018)	Somali parents: purposive sampling (participants were recruited via Autism Independence's social media network)[#1]
	Fox et al. (2017)	
2	**Ami-Narh (2019)**	Ghanaian families and their adolescents with autism: "The sample was recruited from two autism centres in the Greater Accra region of Ghana."
3	**Anderson et al. (2021)**	Parents/guardians: N/A
		Autistic students: "Students were recruited from 14 schools (4 in Florida, 4 in Massachusetts, and 6 in New York)."
4	**Brookman-Frazee (2012)**	BRIDGE (parents): "The eight initial members of the group invited other individuals based on their roles in providing early intervention to this population, in agencies funding services for this population, or as caregivers to a child with ASD, and because they were considered local experts by the community."
		Focus group (parents): N/A
5	**Fletcher-Watson et al. (2017)**	Focus group (parents): N/A
		Piloting survey (parents): N/A
		Survey (parents): Convenience sampling ("The survey was made available online and distributed by researchers affiliated to the COST ESSEA network", "recruitment routes were largely via parents' associations, advocacy groups for autistic adults and professional bodies", "the survey was advertised through a variety of social media and directed to the professional networks of the authors", "in Italy and the United Kingdom, recruitment included circulation of the survey to participants who had previously taken part in early autism research studies (i.e. parents of children with autism), either directly through a register of former participants or indirectly via social media associated with a research group.")
6	**Fong et al. (2021)**	Korean parents: Purposive sampling (from a list given by a Korean organization providing services, information, and advocacy for families of children with developmental disabilities in the community)
7	**Galpin et al. (2017)**	Parents: "All participants were recruited from a government funded, local special school in inner-city London."
8	**Grinker et al. (2012) (SK)**	South Korea focus group (parents): N/A
9	**Grinker et al. (2012) (SA)**	South Africa focus group (parents): N/A
		South Africa interview (parents): N/A

10	**Groba et al. (2021)**	Family and autistic children: N/A
11	**Hallett et al. (2021)**	Parent/carer: N/A
12	**James (2021)**	Survey (American Samoan parents): Convenience sampling
		Interview (American Samoan parents): Invite participants of a workshop related to [autism] at the American Samoa Department of Education that the author was invited to conduct to voluntarily participate in this study.
13	**Linder et al. (2021)**	Parents and young autistic adult: "The individuals identified with relevant experience and expertise in one or more aspects of the project were invited to serve on the stakeholder panel, provide feedback on the proposal, and provide letters of support to submit with the proposal."; "Two ABA service providers were contacted to serve as recruitment sites in the active pilot phase of the project."
14	**Ogourtsova et al. (2021)**	Parent advisory group (parents/caregivers of children with disabilities): Purposeful recruitment techniques of word of mouth and snowball sampling
15	**Pickard et al. (2016)**	Parents: "Providers who worked with families of children with [autism] provided parents with a recruitment flier outlining the goal of the parent focus groups. Parents who voiced interest were able to call, email, or text the primary investigator to express interest in attending."
16	**Pohl et al. (2020)**	Autistic and non-autistic mothers: "The survey was made available online and disseminated via autism-specific support groups and social media pages."
17	**Rabba et al. (2020)**	Parents: Convenience sampling (Through email invitation to autism industry partners and parents from a previous qualitative study)
18	**Smith et al. (2017)**	Parents: Each of the three local school districts (Los Angeles Unified School District, School District of Philadelphia, and Rochester City School District) formed a partnership group.
19	**Strang et al. (2020)**	Parents: N/A
20	**Yingling et al. (2020)**	Parents: Purposive sampling ("The team recruited coparents of children (ages 2–8) diagnosed with ASD within the last year through diagnostic clinics and pediatricians.")

#1: Considering the participant group of Aabe et al. is highly likely to be the same as Ellen Selman et al. and Fox et al., it was assumed that these three studies share the same sampling method despite Aabe et al. did not provide any information on its sampling procedure.

Table 3c

Participant Characteristics (Inclusion Criteria)

	Author (y)	Inclusion criteria
1	**Aabe et al. (2019)**	Somali parents: N/A
	Ellen Selman et al. (2018)	Somali parents: "Parents of children under 16 years' old, a mix of mothers and fathers."
	Fox et al. (2017)	Somali parents: "(1) being a parent to a child under 16 years of age, who has a diagnosis of autism, (2) identifying as a member of the Bristol Somali migrant community."
2	**Ami-Narh (2019)**	Ghanaian families and their adolescents with autism: Receiving services from two autism centres in the Greater Accra region of Ghana
3	**Anderson et al. (2021)**	Parents/guardians: N/A
		Autistic students: "(1) placement in grades, K-8 (2) receipt of individual educational plan (IEP) services under the educational category of Autism in their district; and (3) confirmation of [autism] diagnostic criteria via the Autism Diagnostic Observation Schedule, 2nd Edition as administered by a research-certified diagnostician."
4	**Brookman-Frazee (2012)**	BRIDGE (parents): N/A
		Focus group (parents): N/A
5	**Fletcher-Watson et al. (2017)**	Focus group (parents): N/A
		Piloting survey (parents): N/A
		Survey (parents): N/A
6	**Fong et al. (2021)**	Korean parents: "Parents had a child diagnosed with [autism] and that their racial/ethnic background was Korean; children included in the study had obtained a standardized clinical diagnosis of [autism] from a qualified psychologist, pediatrician, or psychiatrist associated with the provincial government-funded autism assessment network, or through a qualified private clinician."
7	**Galpin et al. (2017)**	Parents: "All respondents with a child(ren) attending the government funded, local special school in inner-city London were invited to take part in a brief survey."
8	**Grinker et al. (2012) (SK)**	South Korea focus group (parents): N/A
9	**Grinker et al. (2012) (SA)**	South Africa focus group (parents): N/A
		South Africa interview (parents): Bilingual Zulu-/English-speaking parents
10	**Groba et al. (2021)**	Family: "Participants were three direct relatives of people with [autism] and, in turn, they were part of the organizations for people with this diagnosis."

		Autistic children: "All participants were selected from a special education centre and met the following criteria: (a) a diagnosis of [autism]; (b) persistent difficulties in daily functioning; and (c) not having used the specific software prior to testing."
11	**Hallett et al. (2021)**	Parent/carer: "Being a parent/carer of a child with [autism] (4–8 years old), with sufficient English to participate."
12	**James (2021)**	Survey (American Samoan parents): "Voluntary stakeholders (i.e., parents, teachers, therapists, community members) with/treating school-age children who have [autism] in the American Samoa School District"
		Interview (American Samoan parents): "Voluntary stakeholders (i.e., parents, teachers, therapists, community members) with/treating school-age children who have an [autism] in the American Samoa School District"
13	**Linder et al. (2021)**	Parents and young autistic adult: "Individuals with practical and personal experience with various areas of the project, specifically, [autism], animal-assisted intervention and Applied Behavioural Analysis."
14	**Ogourtsova et al. (2021)**	Parent advisory group (parents/caregivers of children with disabilities): "Residency in the province of a study site, having a young child (preferably <10y) with a developmental disability who is medically stable (i.e. >1y post-diagnosis or other major medical treatments), for the parent's own health to be stable, having experience dealing with health, educational and social services on behalf of their child in their province, and having access to a computer/ smartphone and be comfortable with applications such as Skype and FaceTime."[#1]
15	**Pickard et al. (2016)**	Parents: "Parents of a child with a current [autism] diagnosis, all parents were Medicaid eligible based on income, and had at least one child between the ages of 18 months and 6 years who was receiving services through the [autism] Medicaid Benefit in Michigan."
16	**Pohl et al. (2020)**	Autistic and non-autistic mothers: "Recruitment was targeted towards mothers who had at least one autistic child, of any age."
17	**Rabba et al. (2020)**	Parents: "Parents who previously received their child's early diagnosis of [autism] (<36 months)."
18	**Smith et al. (2017)**	Parents: "Parents were primary caregivers of one or more children with [autism] in the district."
19	**Strang et al. (2020)**	Parents: "Adolescents age 12-19 years old who met DSM-5 criteria for gender dysphoria and [autism] or social communication disorder (SCD; American Psychiatric Association, 2013) were referred by clinic-based providers in the Gender and Autism Program at Children's National Medical Center to the group program and study if their parents were affirming of their gender needs (as determined from the initial parent interviews)."[#2]
20	**Yingling et al. (2020)**	Parents: "Parents with children recently diagnosed with [autism]."

#1: Although Ogourtsova et al., 2021 only included young children "with a developmental disability", majority of participants engaged were parents of an autistic child(ren) (67%) and autism is one type of developmental disability that affects children. Therefore, it was considered that Ogourtsova et al., 2021 intended to engage participants that had personal experiences with autism, and it met the inclusion criteria of this review.

#2: The age range of autistic adolescents in the inclusion criteria of this study is slight over (by 1 year old) comparing to our inclusion criteria. However, we are focusing on their parents here and the actual age range of the autistic adolescents engaged was not recorded. Therefore, this study was included. This is also the only study that addressed gender diversity out of all the included studies. It could have a big impact on the results of our review if we decided to exclude it because the age range of autistic adolescents is allowed up till 19 years old instead of 18 years old that is what we are searching for. Lastly, the reported mean age and standard deviation were 15.92 and 1.85 (Table 3d), respectively, this means that the participants engaged indeed fit with the inclusion criteria of this review.

Table 3d

Participant Characteristics (Age and Gender of the Autistic Children and Their Parents who were Actively Engaged in the Included Studies)

		Parents of Autistic Child			**Autistic Child**			**Total Number of Participants Responded (n)**
		Age Range (n years old)	**Mean Age (SD[#1])**	**Gender (%male)**	**Age Range (n years old)**	**Mean Age (SD[#1])**	**Gender (%male)**	
1	**Aabe et al. (2019)**	Aabe et al.: N/A	N/A	N/A	N/A	N/A	N/A	15 parents
	Ellen Selman et al. (2018)	N/A	36 (N/A)	20%	4 to 13	7 (N/A)	70.6%	15 parents (of 17 autistic children)
	Fox et al. (2017)							
2	**Ami-Narh (2019)**	Focus groups: 20 to 44	Focus groups: N/A	Focus groups: 50%	Focus groups: 2 to 18	N/A	N/A	2 caregivers
		Individual interviews: 18 to 55	Individual interviews: N/A	Individual interviews: 25%	Individual interviews: 11 to 17	N/A	Individual interviews: 88.9%	8 parents
3	**Anderson et al. (2021)**	Focus group: N/A	Parents: N/A	Parents: N/A	Parents: N/A	Parents: N/A	Parents: N/A	N/A parents
		Randomized controlled trials: N/A	N/A	N/A	N/A	Autistic students (MAAPS group[#2]): 8.07 (2.0)	71%	28 autistic students
						Autistic students (ETAU group[#2]): 8.14 (2.1)	79%	

4	**Brookman-Frazee (2012)**	BRIDGE: N/A	N/A	N/A	N/A	N/A	N/A	3 parents
		Focus groups: N/A	N/A	N/A	N/A	N/A	N/A	10 parents
5	**Fletcher-Watson et al. (2017)**	Focus group (parents): N/A	N/A	N/A	N/A	N/A	N/A	N/A parents
		Piloting survey (parents): N/A	N/A	N/A	N/A	N/A	N/A	3 parents
		Survey (parents): N/A	Survey: 41 (8.4)	Survey: 14%	N/A	N/A	N/A	1040 parents
6	**Fong et al. (2021)**	39 to 59	48.10 (13.02)	15%	N/A	N/A	N/A	20 parents
7	**Galpin et al. (2017)**	Survey: N/A	N/A	22.3%	4.3 to 18.7	N/A	82%	139 parents
		Semi-structed interview: N/A	N/A	11.80%	5.5 to 17.6	10.6 (3.7)	88.8%	17 parents[#3]
8	**Grinker et al. (2012) (SK)**	Focus group: N/A	N/A	N/A	N/A	N/A	N/A	16 parents
9	**Grinker et al. (2012) (SA)**	Focus group: N/A	N/A	N/A	N/A	N/A	N/A	N/A parents
		Interview: N/A	N/A	N/A	1.5 to 3	N/A	N/A	N/A parents
10	**Groba et al. (2021)**	N/A	57 (4)	0%	N/A	N/A	N/A	3 family members
		N/A	N/A	N/A	10-13	12 (1)	33.3%	3 children
11	**Hallett et al. (2021)**	Intervention: N/A	N/A	N/A	Verbal group: 6.6 to 8.8	Verbal group: 7.5	91.6%	12 parents/carers
					Minimally verbal group: 4.1 to 9.2	Minimally verbal group: 7		

		Public and Patient Involvement Panels: N/A	N/A	N/A	N/A	N/A	N/A	2 parents/carers
		Feasibility study: N/A	N/A	N/A	N/A	N/A	N/A	24 parents/carers
		Semi-structured telephone interviews: N/A	N/A	N/A	N/A	N/A	N/A	9 parents/carers
12	**James (2021)**	Survey: N/A	N/A	N/A	N/A	N/A	N/A	65 parents
		Interview: N/A	N/A	N/A	N/A	N/A	N/A	11 parents
13	**Linder et al. (2021)**	N/A	N/A	N/A	N/A	N/A	N/A	3 parents
		N/A	N/A	N/A	N/A	N/A	N/A	1 young autistic adults
14	**Ogourtsova et al. (2021)**	N/A	N/A	16.7%	N/A	N/A	N/A	6 parents/ caregivers
15	**Pickard et al. (2016)**	N/A	34.81 (7.07)	12.5%	1.5 to 6	4.50 (0.73)	77.8%	16 parents
16	**Pohl et al. (2020)**	N/A	42.7 (8.4)	<1%[#4]	N/A	N/A	N/A	335 autistic mothers
		N/A	44.6 (9.1)	2%[#4]	N/A	N/A	N/A	132 non-autistic mothers
17	**Rabba et al. (2020)**	42 to 54	N/A	33.3%	1.4 to 2.6	N/A	66.7%	3 parents
18	**Smith et al. (2017)**	N/A	N/A	N/A	N/A	N/A	N/A	N/A parents
19	**Strang et al. (2020)**	Group sessions: N/A	N/A	37.8%	N/A (adolescents)	15.92 (1.85)	N/A[#5]	18 families (33 parents)[#6]
		Usefulness ratings: N/A						29 families[#6]

20	**Yingling et al. (2020)**	21 to 68	N/A	36.1%[#7]	N/A	N/A	72.1%[#7]	61 parents

Note: The data were divided by each section of the study in the 20 settings. One setting referred to one type of intervention (for example, focus group, interview, randomized controlled trials, panel discussion, and survey) was used in a section of the included research, which was represented by one row in this table. Each section could include the same or different types of research participant as other sections in the same research.

#1: Standard deviation (SD)

#2: Modular approach for autism programming in schools (MAAPS) group; enhanced treatment as usual (ETAU) group

#3: Those who participated in the semi-structured interview is a subgroup of parents selected from those who participating in the survey

#4: Information on the participants' sex and gender were included. For autistic mothers' sex, % of "female (sex)" were 99% and % of "male (sex)" were <1. Autistic mothers' gender were 95% "female (gender)", <1% "male (gender)" and 4% "others (gender)". Among non-autistic mothers, 100% were "female (sex)". 98% of non-autistic mothers were "female (gender)" and 2% were "male (gender)".

#5: It was described by Strang et al., 2020 that 16 binary-trans-females (assumed male at birth), 11 binary-trans-males (assumed female at birth), and four nonbinary-transgenders (assumed female at birth) were engaged at the beginning of the study. By the end of the study, gender had been fluid for four participants that two moved from binary to nonbinary identities, one from binary-transgender to cisgender, and one from nonbinary-transgender to cisgender.

#6: Strang et al., 2020 provided information in terms of the number of families involved, as opposed to the number of parents involved that was how other included studies presented. In Strang et al., 2020, 33 parents from 18 families were involved in the "group sessions" section. In the "usefulness ratings" section, we only knew that 11 additional families (adding to the 18 families from "group sessions") were involved. We do not know the exact number of parents from these 11 additional families that were involved. Consequently, when counting the total number of parents involved in Strang et al., 2020, we assumed at least one family members of the 11 families were involved in "usefulness ratings". We were aware that there could be more than one family members from these 11 families were involved, thus we concluded that there were at least 44 parents (33 parents from "group sessions" plus at least 11 parents from "usefulness ratings") involved in both sections of Strang et al., 2020

#7: It was reported that there were some missing information on parents gender demographics (n = 2, 3.3%) and missing child gender (n = 4, 6.6.%).

Table 3e

Calculated Average Age, Mean Age and % of Male Autistic Children and Their Parents

	Parents	**Autistic children**
Calculated average age (years)[#1]	44	N/A[#3]
Calculated mean age (years)[#2]	43.5	8.6
Calculated % of male	21.6	87.6

#1: Calculation was made based on the age range available in Table 3d (dividing the differences between the maximum and minimum age range by two).
#2: Calculations were made based on the mean age available in Table 3d (dividing the sum of all recorded mean age by the total number of participant groups that reported this information in the included studies).
#3: The calculations were not applicable because there were participant groups with a wide age range that covers the age range of the entire inclusion criteria of this review (Ami-Narh, 2019) and some with relatively narrow age range (6.6. to 8.8 years old) (Hallett et al., 2021). Therefore, it might not be a helpful information to calculate the average age of autistic children engaged based on the maximum and minimum of "age group" data.

Table 4

Strengths and Limitations in Participant Engagement

		Participant Engagement Strengths	Participant Engagement Limitations
1	**Aabe et al. (2019)**	N/A	N/A
2	**Ami-Narh (2019)**	• "…, the input from the community, including stakeholders (practitioners, administrators) as well as end-users (families of children with autism) allowed the revision and adaptation of an existing evidence-based intervention to be developed that is culturally responsive to the Ghanaian families, who currently has no formal services available."	• "…, number of participants in the sample was restricted and may not be representative of caregivers and practitioners in Ghana…" • "… because participants were specifically targeted based on their affiliation to the autism centers, participants may have been biased while participating in the study."
3	**Anderson et al. (2021)**	N/A	• "… the study was designed for a small N; thus, it was underpowered." • "… it will be critical for such evaluations to include student participants with diversity in presentation of cognitive levels and characteristics to help identify whether the [Modular Approach for Autism Programs in Schools] framework has the sufficient flexibility to be applicable for any student with [autism] or is more effective for students with specific needs."
4	**Brookman-Frazee (2012)**	N/A	• "… it is not known if this model is feasible in other geographic locations that have different structures for community early intervention programs and may not have local services researchers with whom to partner."
5	**Ellen Selman et al. (2018)**	N/A	• "We chose to recruit a relatively small sample, which reflected our exploratory aim of achieving insight into this seldom heard group, and enabled in-depth analysis." • "… we interviewed only three men."
6	**Fletcher-Watson et al. (2017)**	N/A	• "The limitations of [convenience sampling] mean that the final sample may not be representative of the wider community…" • "… it is possible that the very positive attitudes to research observed would not be shared by a sample from a less research-engaged background…"

7	**Fong et al. (2021)**	N/A	• "… small sample size…" • "… the majority of respondents being mothers may limit the generalizability of findings." • "… the recruitment of Korean parents who were contacted from a local, non-profit organization in the community may not represent the views and experiences of parents who may be less connected with their communities."
8	**Fox et al. (2017)**	• "The community-based participatory approach ensured that the design and conduct of the research was, at all stages, culturally sensitive and responsive to the needs of the local Somali community."	• "This was a small scale, exploratory qualitative study and as such, the findings should be considered as an insight into the views and experiences of a Somali Community in one UK city, …" • "It should be noted that the first nine parents all volunteered to participate and were therefore self-selecting and were engaged with [Autism Independence] and thus had more access to information and support from other parents raising a child with autism." • "Our sample included families with children aged 4–14 years of age…, it did preclude exploration of issues pertinent to older children."
9	**Galpin et al. (2017)**	N/A	• "… the fact that the research was conducted by school employees might have affected the results in some way."
10	**Grinker et al. (2012)**	N/A	N/A
11	**Groba et al. (2021)**	N/A	• "… the small number of participants in the group of children with autism and relatives." • "Difficulties in recruiting more participants in the children's group were related to the difficulty in [engaging] minors in research studies due to ethical considerations and the difficulties in conducting participatory studies in children with [autism] with persistent communication difficulties." • "Non-participation of adults with autism as a participants' group is considered a limitation"
12	**Hallett et al. (2021)**	N/A	• "… the [Public and Patient Involvement] panel was small, and weighted towards adults with [autism] rather than parents."

			• "It would have been helpful to have a greater number of parents [engaged] in the initial concept development." • "The participant sample size was also small and quantitative parent and child outcomes have not yet been explored in comparison to a control condition."
13	**James (2021)**	• "The research methodology used… will be highly generalizable to other [culturally diverse] populations."	• "… the culturally adapted intervention is likely not generalizable to other diverse groups of students with autism due to it utilizing factors specific to American Samoan culture. The process would need to be completed with the new population to determine the cultural relevance of the intervention."
14	**Linder et al. (2021)**	N/A	N/A
15	**Ogourtsova et al. (2021)**	N/A	• "We were limited by the small sample size in both study populations." • "To address the homogeneity of the [parent advisory group]/lack of diversity, we suggest recruiting additional members with diverse characteristics. This might be established by broadening our current recruitment strategies to reach and attract fathers of disabled children." • "All co-authors were participants in this study, which might have introduced a certain response bias"
16	**Pickard et al. (2016)**	• "… parent and provider focus groups were run separately. The primary reason for this separation was due to suggestions from our community partners that combined focus groups might result in power dynamics that would make it more difficult for parents to express their experience and opinions."	• "The study results are drawn from a small number of focus groups and participants from within a specific community setting. Therefore, the results may not generalize to other low-resourced parents outside of the metro-Detroit area."
17	**Pohl et al. (2020)**	N/A	• "… our sample of women may not be representative of both the general and autistic population of mothers and therefore may reduce the generalisability of our findings." • "Our non-autistic sample may not be representative of the general population of mothers. Our non-autistic sample only included mothers with at least one autistic child and included a higher than

			usual proportion of women who had experienced postpartum depression." • "… mothers in our samples were also from predominantly Western countries, suggesting that the themes reported here may not be applicable to women from non-Western countries." • "… only mothers who were literate, able to understand our questions and with access to a computer were able to complete the survey, again highlighting that the results from this survey may not be representative of all autistic mothers in the population." • "Future studies should include a neurotypical non-autistic group." • "… the average of age of children and mothers in our study was quite high, with children being adolescents and mothers about 40 years of age at the time of completion of the survey."
18	**Rabba et al. (2020)**	N/A	• "Engaging a larger sample of parents whose child had previously been diagnosed with autism proved to be a significant challenge." • "As the website has not been translated into other languages, limiting the population it can benefit to those who only understand English."
19	**Smith et al. (2017)**	N/A	• "District administrators identified potential partnership members. Doing so ensured that partnership members had good working relationships with the district, but it may have led to the underrepresentation of some stakeholder groups, such as teachers."
20	**Strang et al. (2020)**	• "Including the complementary perspectives of parents *and* youth was central to this study's design to maximize the relevance of the clinical care model…"	• "there may be intrinsic bias to [utility ratings] given that 18 of the 29 youth who provided final utility responses were part of the qualitative needs assessment group that drove the development of the curriculum."
21	**Yingling et al. (2020)**	N/A	N/A

Table 5a

Facilitators of Patient Engagement in Research

Inequalities	Stakeholders	Facilitators	Aabe et al. (2019)	Ami-Narh (2019)	Anderson et al. (2021)	Brookman-Frazee (2012)	Ellen Selman et al. (2018)	Fletcher-Watson et al. (2017)	Fong et al. (2021)	Fox et al. (2017)	Galpin et al. (2017)	Grinker et al. (2012)	Groba et al. (2021)	Hallett et al. (2021)	James (2021)	Linder et al. (2021)	Ogourtsova et al. (2021)	Pickard et al. (2016)	Pohl et al. (2020)	Rabba et al. (2020)	Smith et al. (2017)	Strang et al. (2020)	Yingling et al. (2020)
Access	**Community organizations**	Community initiating the research	Y			Y						Y			Y				Y				
		Co-development/ co-design of the research	Y			Y	Y		Y		Y			Y			Y	Y	Y		Y		Y
	Researchers	Increasing accessibility/ accommodations made for data collection	Y			Y			Y	Y	Y			Y	Y			Y		Y			Y
		Modifying and adapting materials via participant input		Y	Y								Y						Y	Y		Y	
	Funders	Participant/ stakeholder engaged in funding process				Y										Y							

Inequalities	Stakeholders	Facilitators	Aabe et al. (2019)	Ami-Narh (2019)	Anderson et al. (2021)	Brookman-Frazee (2012)	Ellen Selman et al. (2018)	Fletcher-Watson et al. (2017)	Fong et al. (2021)	Fox et al. (2017)	Galpin et al. (2017)	Grinker et al. (2012)	Groba et al. (2021)	Hallett et al. (2021)	James (2021)	Linder et al. (2021)	Ogourtsova et al. (2021)	Pickard et al. (2016)	Pohl et al. (2020)	Rabba et al. (2020)	Smith et al. (2017)	Strang et al. (2020)	Yingling et al. (2020)
Information	**Community organizations**	Acknowledging and incorporating diverse viewpoints	Y													Y							
	Researchers																						
	Funders	N/A																					
Validity	**Community organization/ researchers**	Inclusion of community / participant voices in research development	Y	Y		Y	Y	Y	Y		Y	Y		Y	Y	Y	Y						
		Inclusion of community / participant voices in the interpretation of results			Y	Y												Y	Y			Y	
	Funders	N/A																					
Ownership	**Community organization/ researchers**	Inclusion of participants/ collaborators in the research process	Y			Y									Y								
	Funders	N/A																					

Inequal-ities	Stakeholders	Facilitators	Aabe et al. (2019)	Ami-Narh (2019)	Anderson et al. (2021)	Brookman-Frazee (2012)	Ellen Selman et al. (2018)	Fletcher-Watson et al. (2017)	Fong et al. (2021)	Fox et al. (2017)	Galpin et al. (2017)	Grinker et al. (2012)	Groba et al. (2021)	Hallett et al. (2021)	James (2021)	Linder et al. (2021)	Ogourtsova et al. (2021)	Pickard et al. (2016)	Pohl et al. (2020)	Rabba et al. (2020)	Smith et al. (2017)	Strang et al. (2020)	Yingling et al. (2020)
Value	**Community organization/ researchers**	Building longstanding relationships beyond the project	Y									Y											
		Adapting research content to produce value for community organizations		Y	Y											Y		Y					Y
	Funders	Participants as recipients of funding													Y						Y		
Account-ability	**Community organization/ researchers**	Building trust-based relationship	Y		Y							Y					Y				Y		
		Acknowledging roles of power/ privilege/risk		Y		Y									Y		Y	Y					Y
	Funders	Engaging in funding application(s)																			Y		
Author-ship	**Community organization/ researchers**	Stakeholders/ collaborators engaged in authorship of results					Y								Y	Y						Y	
	Funders	N/A																					

Table 5b

Barriers of Patient Engagement in Research

Inequalities	Stakeholders	Barriers	Aabe et al. (2019)	Ami-Narh (2019)	Anderson et al. (2021)	Brookman-Frazee (2012)	Ellen Selman et al. (2018)	Fletcher-Watson et al. (2017)	Fong et al. (2021)	Fox et al. (2017)	Galpin et al. (2017)	Grinker et al. (2012)	Groba et al. (2021)	Hallett et al. (2021)	James (2021)	Linder et al. (2021)	Ogourtsova et al. (2021)	Pickard et al. (2016)	Pohl et al. (2020)	Rabba et al. (2020)	Smith et al. (2017)	Strang et al. (2020)	Yingling et al. (2020)
Access	**Community organizations**	Lack of organizational resources			Y																		
		Impediment to diverse participants		Y													Y	Y					
		Local contextual impediments										Y											
	Researchers	Roles as insider and outsider	Y					Y															
	Funders	N/A																					
Information	**Community organizations**	Lack of understanding the research process	Y																				
	Researchers	N/A																					
	Funders	N/A																					
Validity	**Community organizations**	Lack of community review and input		Y																			
	Researchers	N/A																					
	Funders	N/A																					

Inequalities	Stakeholders	Barriers	Aabe et al. (2019)	Ami-Narh (2019)	Anderson et al. (2021)	Brookman-Frazee (2012)	Ellen Selman et al. (2018)	Fletcher-Watson et al. (2017)	Fong et al. (2021)	Fox et al. (2017)	Galpin et al. (2017)	Grinker et al. (2012)	Groba et al. (2021)	Hallett et al. (2021)	James (2021)	Linder et al. (2021)	Ogourtsova et al. (2021)	Pickard et al. (2016)	Pohl et al. (2020)	Rabba et al. (2020)	Smith et al. (2017)	Strang et al. (2020)	Yingling et al. (2020)
Ownership	**Community organizations**	N/A																					
	Researchers																						
	Funders																						
Value	**Community organizations**	Stigma in cultural conceptions of autism					Y					Y											
	Researchers	N/A																					
	Funders	N/A																					
Accountability	**Community organizations/ researchers**	Lack of trust and comfort										Y											
	Funders	N/A																					
Authorship	**Community organizations**	N/A																					
	Researchers																						
	Funders																						

2.9 Appendices

Appendix A
Ovid Medline Search Strategy

Ovid MEDLINE(R) and Epub Ahead of Print, In-Process, In-Data-Review & Other Non-Indexed Citations, Daily and Versions(R) <1946 to February 22, 2022>

1. exp Autism Spectrum Disorder/
2. exp Asperger Syndrome/
3. exp Autistic Disorder/
4. autism.mp.
5. 1 or 2 or 3 or 4
6. exp Child/
7. child*.mp.
8. exp Adolescent/
9. youth*.mp.
10. teen*.mp.
11. exp Young Adult/
12. exp Infant/
13. exp Child, Preschool/
14. exp Pediatrics/
15. exp Parents/
16. exp Legal Guardians/
17. exp Caregivers/
18. 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17
19. exp Patient Participation/
20. patient involvement.mp.
21. patient engagement.mp.
22. 22 ((particip* or stakeholder* or community* or patient) adj3 (research or engage*)).mp.
23. 19 or 20 or 21 or 22
24. 5 and 18 and 23
25. limit 24 to (yr="2011 - 2021" and english)

Appendix B

Ovid Embase Search Strategy

Embase <1974 to 2022 February 22>

1. exp Autism Spectrum Disorder/
2. exp Asperger Syndrome/
3. exp Autistic Disorder/
4. autism.mp.
5. 1 or 2 or 3 or 4
6. exp Child/
7. child*.mp.
8. exp Adolescent/
9. youth*.mp.
10. teen*.mp.
11. exp Young Adult/
12. exp Infant/
13. exp Child, Preschool/
14. exp Pediatrics/
15. exp Parents/
16. exp Legal Guardians/
17. exp Caregivers/
18. 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17
19. exp Patient Participation/
20. patient involvement.mp.
21. patient engagement.mp.
22. ((particip* or stakeholder* or community* or patient) adj3 (research or engage*)).mp.
23. 19 or 20 or 21 or 22
24. 5 and 18 and 23
25. limit 24 to (yr="2011 - 2021" and english)

Appendix C

Web of Science Core Collection Search Strategy

Web of Science Core Collection

1. autism (All Fields)
2. "autism spectrum disorder" (All Fields)
3. asperger syndrome (All Fields)
4. autistic disorder (All Fields)
5. 1 or 2 or 3 or 4
6. child* (All Fields)
7. adolescent* (All Fields)
8. youth* (All Fields)
9. teen* (All Fields)
10. "young adult*" (All Fields)
11. infant* (All Fields)
12. preschool* (All Fields)
13. p$ediatric* (All Fields)
14. parent* (All Fields)
15. guardian* (All Fields)
16. caregiver* (All Fields)
17. 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16
18. patient participation (All Fields)
19. patient involvement (All Fields)
20. patient engagement (All Fields)
21. 18 or 19 or 20
22. 5 and 17 and 21
23. Limit 23 to Publication Years 2021 to 2011and English (Languages)

Appendix D

Ovid APA PsycINFO Search Strategy

APA PsycInfo <1987 to February Week 2 2022>

1. exp Autism Spectrum Disorders/
2. autism.mp.
3. 1 or 2
4. exp Child Health/
5. child*.mp.
6. exp Adolescent Health/
7. exp Family/
8. teen*.mp.
9. youth*.mp.
10. young adult*.mp.
11. exp Pediatrics/
12. parent*.mp.
13. exp Parents/
14. exp Caregivers/
15. exp Child Care/
16. adolescent*.mp.
17. infant*.mp.
18. exp Preschool Students/
19. exp High School Students/
20. exp Primary School Students/
21. exp Nursery School Students/
22. exp Kindergarten Students/
23. 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22
24. exp patient participation/
25. patient involvement.mp.
26. exp Patient Centered Care/
27. patient engagement.mp.
28. ((particip* or stakeholder* or community* or patient) adj3 (research or engage*)).mp.
29. 24 or 25 or 26 or 27 or 28
30. 3 and 23 and 29
31. limit 30 to (english language and yr="2011 - 2021")

Appendix E
EBSCOhost CINAHL Search Strategy

1. MH "Autistic Disorder"
2. MH "Asperger Syndrome"
3. MH "Pervasive Developmental Disorder-Not Otherwise Specified"
4. 1 or 2 or 3
5. MH "Adolescence"
6. MH "Child"
7. MH "Parents"
8. MH "Child, Preschool"
9. MH "Infant"
10. MH "Child Health"
11. MH "Adolescent Health"
12. MH "Caregivers"
13. MH "Guardianship, Legal"
14. 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13
15. MH "Stakeholder Participation"
16. MH "Family Involvement (Iowa NIC)"
17. MH "Decision Making, Shared"
18. MH "Participation: Health Care Decisions (Iowa NOC)"
19. MH "patient involvement"
20. MH "patient participation"
21. MH "patient empowerment"
22. MH "community engagement"
23. MH "Family Centered Care"
24. MH "Patient Centered Care"
25. MH "patient empowerment"
26. 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25
27. 4 and 14 and 26
28. Limit 27 to Published Date: 20110101-20211231 AND Language: English

Appendix F
Participant Characteristics (Raw Data)

https://macdrive.mcmaster.ca/f/2e0eb3dbcb744acb972a/?dl=1

Appendix G

Facilitators of Patient Engagement in Research (Raw Data)

https://macdrive.mcmaster.ca/f/933224a7e6f04c7b87c7/?dl=1

Appendix H

Barriers of Patient Engagement in Research (Raw Data)

https://macdrive.mcmaster.ca/f/42b302df914b4d80bba4/?dl=1

Chapter 3: Conclusion

3.1 Summary of Findings

In conclusion, the scoping review suggests that marginalized populations, including autistic children, were under-represented in autistic research. Some of the identified under-represented marginalized autistic children populations were those (1) those living in developing or under-developed countries, (2) those who received autism services from treatment centres and schools/specific education centres that do not have existing connections to collaborate with researchers, (3) families of ethnic minority in Western countries, (4) autistic children who received their diagnosis later in their childhood, (5) families whose first language is not English, (6) male parents of autistic children, (7) female autistic children, (8) families with low household income who are not enrolled in governmental healthcare financial support program and (9) those who do not have access to technological devices or lack technological literacy skills. The identified facilitators of patient engagement in autistic children's research were: (1) building longstanding, trust-based relationships among community organizations, researchers and funders, (2) engaging patients in all stages of research development, and (3) participants engagement in research funding. The identified barriers were: (1) allocation of research funding, (2) identity conflict, lack of professional knowledge, (3) design of inclusion/exclusion criteria and sampling methods; applicability of research evidence, and (4) social stigmatization towards autism.

3.2 Implications

3.2.1 Language in Discussing Autism

Most of the research still refers autism as "autism spectrum disorder". This is understandable because this is how the fifth version of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) named autism (American Psychiatric Association, 2013). In

recent years (after the publication of DSM-5 in 2013), there have been discussions about whether it is better to call autism as a condition instead of a disorder. This is to minimize the social stigma the word "disorder" contributes to autism. I expect that the next version of DSM might address this change. I chose to refer autism as a condition to promote an equity-based approach to autism research.

3.2.2 Imbalanced Sex Ratio Population in Autism Research

There were obvious differences between the proportion of male and female autistic individual populations engaged in the scoping review. Most of the parents engaged were female while most of the autistic children engaged were male. This is likely due to the social recognition of female or mother usually takes up most of the responsibility of taking care of their child. However, as reminded by Pohl et al., 2020, a mother does not limit to one's gender and being a female is not a must to take up the identity as a "mother'. Gender equity can be promoted by equally engaging male and female parents of an autistic child in autism research. The dominancy of male autistic children engaged is likely due to the four times higher prevalence of autism in males compared to females (Beggiato et al., 2017; Public Health Agency of Canada [PHAC], 2016). Autism is underestimated and often misdiagnosed in females (Haney, 2016). The diagnostic guidelines were designed in reference to the symptoms, especially restricted and repetitive behaviours, that are commonly observed among autistic males (Werling & Geschwind, 2013). This poses challenges for health professionals to diagnose autism among females. As a result, the imbalanced proportion of autism diagnosis made among males and females might make it easier for researchers to engage male autistic children compared to female autistic children. This imbalanced patient engagement needs to be improved. An effort needs to be made to engage more female autistic children in research.

3.2.3 Lack of Studies Conducted in Canada

Most of the included studies from the review were conducted in high-income, Western countries. Whilst Canada is typically on the list considered as one of the high-income, Western countries, it conducted relatively fewer studies in this field as compared to other countries. Specifically, the United States (US) (n = 9, 45%) and the United Kingdom (UK) (n = 5, 25%) were where most of the included studies were conducted. This shows that more patient engagement studies investigating autistic children and their parents are needed to be conducted in the Canadian context. This is something that Canadian policymakers should encourage. This raises the concern of which kind of patient-oriented research (POR) evidence the healthcare decision related to autistic children in Canada were based on. Since there were more research data available on this topic from the US and the UK, I expect that the Canadian health policymakers might have been relying on POR data produced from these countries to make the decisions in Canada. However, this might not be appropriate since each society has a different structure of healthcare system that can affect its implementation of patient engagement in research.

The lack of studies conducted in Canada (n = 3, 15%) contributes to a major limitation to attempting to apply these recommendations to the Canadian context. Since the recommendations made in the scoping review were derived based on the data extracted from the included studies, thus there is a concern as to whether these recommendations are applicable in the Canadian context. However, we still discuss what the implications of these recommendations mean in Canada because most of the included studies were conducted in high-income, Western countries. Canada is typically considered in the list as one of the high-income, Western countries around the globe. Most importantly, there is an urgent need for Canadians to start developing research evidence on patient engagement initiatives with autistic children in the local context.

3.2.4 Implications of Patient Engagement Recommendations in Local Context

Based on the patient engagement recommendations generated from the scoping review, I explored what the patient engagement recommendations mean to our local policymakers, that is to our Canadian healthcare system. In Canada, the federal and provincial government both plays a role in the healthcare system. Therefore, here, we are specifying the implementation of these recommendations at Hamilton in the province of Ontario, which is where our research team locates.

Defined by the Canadian Institutes of Health Research (CIHR), the practice of knowledge translation (KT) is "a dynamic and iterative process that includes synthesis, dissemination, exchange, and ethically sound application of knowledge to improve the health of Canadians" (Canadian Institutes of Health Research Government of Canada, 2005). CIHR is one of the major federal research grant organizations in Canada. KT has the potential to narrow the gaps between research evidence and health decision-making, thus providing more effective evidence-informed healthcare for Canadians. KT strategies are the strategy that can be used to facilitate the implementation of an intervention, practice, or innovation (Armstrong, 2013). By matching the recommendations generated from facilitators and barriers to KT strategies, it can help to increase the chance of success in improving patient engagement in research. Support from KT specialists could also be helpful to facilitate this KT process. Below, I listed the details of some of the resources that can be used in Hamilton, Ontario to implement the recommendations that we made from the scoping review.

Firstly, we suggested engaging patients in all stages of research development can improve patient engagement in autistic children's research; this includes before, during and after the research study is conducted (Sacristán, 2016). Particularly, of those included studies in the scoping review, there was a lack of engagement of autistic children and their parents before the study. Identifying research priorities and leading and designing research are some

of the key events for patient engagement in the stage of before research (Sacristán, 2016). Researchers should be encouraged to engage patients in the production of research protocol to the local research ethics board (REB), that is Hamilton Integrated Research Ethics Board (HiREB), for ethics approval or to CIHR for federal research grant applications. It is not only to obtain their feedback on a research protocol written by the researchers but since the initiation of the research project based on their research priorities. As researchers and community's research priorities are typically different, it is expected that there will be conflicts among the community members, researchers and funders' research priorities (Crowe et al., 2015). It is important to remind researchers to be open-minded, respect the core of POR and prioritize community members' priorities whenever possible.

Secondly, building longstanding, trust-based relationships among community organizations, researchers and funders are vital in allowing participants to actively engage in research. Addressing power dynamics is needed to build positive relationships among these stakeholders, so as to build a sense of belonging for all parties engaging in research. Researchers need to be patient with developing a positive rapport in research-participant relationships because this takes time, and it is challenging to maintain through the course of the research study (Pitts, 2007). To build a partnership, interpersonal connections between the researcher and participant need to be achieved (Pitts, 2007). Referencing the different influencers and dimensions of trust can be helpful to effectively enhance trust, despite working with limited time and resources in research (Wilkins, 2018). The dissemination of research findings is essential to close the evidence-policy gap. The role of funders in research should not be "pay and leave". Community organizations, researchers and funders should be actively engaged in this stage of research to maximize the use of evidence in policy. Government agencies, for example, the Public Health Agency of Canada (PHAC) and Health Canada, can facilitate KT development in research by supporting funding agencies, like CIHR, to fund KT

activities. Some of the procedures that CIHR can do to maximize the use of evidence in health practice and policy are to require a KT plan for funded projects and include KT costs as eligible expenditures (Holmes, 2012).

Thirdly, participant engagement in funding needs to be enhanced to improve patient engagement in research. Funding discussion panels, for example, CIHR in Canada, can always include community members in making the decisions on research grant applications. In Canada, the Tri-Council Policy Statement (TCPS2) is an ethical guideline for research engaging humans. There are no signs that TCPS2 provides any suggestions on the engagement of participants when applying for research funding (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Chapter Seven of TCPS2 ("Conflicts of Interest") discusses situations when conflicts of interest exist among institutions, REB members and researchers (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Based on the results from the scoping review, the role of funding to support the research is rarely described. I would recommend journals to encourage the description of how funding supported the research, rather than only claiming whether there were any conflicts of interest in the study. Dual roles of participant and researcher commonly take place in POR because of its value to empower community members as researchers to co-produce the research. Carrying dual roles can create conflicts of interest. Researchers should take on the responsibility to disclose the nature of this conflict to the participant during the process of obtaining their consent to engage in the study. Researchers can actively support these participants on how they can manage the conflict. The conflict encountered by these participants should also be described when reporting the research findings.

Fourthly, research that is restricted to a local context usually lacks diverse participants. This raises the concern about the applicability of the research results. The design of inclusion criteria and exclusion criteria dictates the sampling method of the study and has a direct impact

on the generalizability of the results. Strict study criteria limit the number and diversity of eligible participants to be engaged in research, which decreases the applicability of research evidence (Khan, 2005). Therefore, researchers and funders have the responsibility to ensure that the study criteria chosen by them pose equitable distribution of the burdens and benefits of research, to ensure justice in research. In chapter 4 of TCPS2 (Fairness and Equity in Research Participation), guidance regarding "appropriate inclusion" and "inappropriate exclusion" was stated (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). The importance of being inclusive when selecting participants is emphasized (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Researchers have the duty to ensure that they do not exclude individuals or groups from participating in research for reasons that are unrelated to the research objective (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). For instance, the Indigenous people and First Nations people is a minority group in Canadian research that requires more acknowledgement (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Researchers should also avoid unfairly including certain groups in research because of convenience that takes advantage of their vulnerability (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). For example, attracting people with limited financial resources to engage in research with financial incentives (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). Children are one of the groups that are listed as frequently inappropriately excluded in research (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). The scoping review I conducted verified this in autism research. TCPS2 believes that the inclusion of children in research is often avoided by researchers because of the challenges in research design arises from their developmental stage, the difficulty of getting their consent because of children's lack of decision-making capacity and uncertain long-lasting physical or psychological harm a child may experience from

engaging in research (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). TCPS2's standard on when to engage children in research was "researchers should not exclude children from research unless there is a valid reason for doing so" and "participation of children in research is justifiable when the research objective cannot be achieved with adult participants only" (Interagency Advisory Panel on Research Ethics Government of Canada, 2019). These two statements can be confusing to understand. Clarifications of these statements of TCPS2 are needed for researchers to ensure an equitable approach to engaging children in research is needed. Providing examples on TCPS2 for different scenarios would be helpful for researchers, REB and regulators to refer to when making relevant decisions. To facilitate the engagement of children with Canadian researchers, the current TCPS2 guideline asks researchers to adhere to the following instructions: "the research question can be addressed only with participants within the identified group; and the research does not expose the participants to more than minimal risk without the prospect of direct benefits for them; or where the research entails only minimal risk, it should at least have the prospect of providing benefits to participants or to a group that is the focus of the research and to which the participants belong" (Interagency Advisory Panel on Research Ethics Government of Canada, 2019).

Moreover, 16% of Hamilton residents live in low-income households, which is a higher proportion than provincial and national averages. Almost one-quarter of them are children under six years of age (Statistics Canada, 2017). Therefore, when making population health decisions, local public health officials at Hamilton Public Health Services should make use of research conducted locally in Hamilton. When there is not enough evidence produced in Hamilton to support the decision-making, Hamilton policymakers can take reference of research conducted in a location that has similar population characteristics as Hamilton, preferably within Canada. Hamilton Public Health Services should also support local researchers to engage children from low-income households in research. This is not only to

promote an equitable approach to conducting research but also to produce research evidence that is applicable in supporting local policymakers to make decisions for Hamilton citizens. Policymakers should avoid generalizing research evidence that was conducted with a specific group to make decisions for the entire population, and vice versa.

Fifthly, the identity conflicts that community members experience from being a researcher have an impact on the participant–researcher dynamics. The lack of knowledge of how research is conducted is one of the barriers for patients to engage in research. Training and education for "expert patients" should be provided. To prepare patients to be "experts" in research, investment in their education is needed. Although CIHR has invested approximately $1.9 billion which is over 19% of its annual investments to support the training of future researchers, there is a lack of expert training course that is designed to teach Canadian patients or their family members about how to conduct research (Canadian Institutes of Health Research Government of Canada, 2016). Expert courses are needed to empower patients to allow them to directly collaborate in research as co-researchers. The Family Engagement in Research Course is one of the few available resources in the field for Canadian researchers and families who have an interest in child neurodevelopmental research (CanChild, 2022). However, this course requires a 500 Canadian Dollars fee upon acceptance to the program (CanChild, 2022). Although there is limited full scholarship, applicants are required to indicate that they do not have funding from any alternative source to apply for the scholarship (CanChild, 2022). The accessibility to this program limits the number and diversity of patients that can receive research training. There is also a need to develop training courses that are specifically targeting patients and their families. One of the expert courses that CIHR can reference is the European Patients' Academy on Therapeutic Innovation Open Classroom (EUPATI). EUPATI is a 14-month in-depth, open-access course for patient advocates that is available in seven languages (English, German, Spanish, Polish, French, Russian and Italian) (European Patients' Academy

on Therapeutic Innovation, n.d.). The six modules of EUPATI cover "discovery of medicines & planning of medicines", "non-clinical testing and pharmaceutical development", "exploratory and confirmatory clinical development", "clinical trials", "regulatory affairs, medicinal product safety, pharmacovigilance and pharmacoepidemiology", and "health technology assessment" (European Patients' Academy on Therapeutic Innovation, n.d.).

Sixthly, the lack of resources to support community organizations engaged in research was a concern. Funders, like the CIHR, should consider allocating more funding to community organizations to facilitate their engagement in research. To ensure that funding can increase community members' engagement in research, CIHR grant applications can add a section to require a detailed plan of engagement with community organization(s) in the research. When releasing the research funding, the required funding to engage these community members can be directly distributed to the community organization(s). This can avoid the lack of input to research from community organizations due to insufficient resources.

Lastly, work needs to be done to work toward removing the stigmatization of autism in the Canadian society to increase participants' trust and comfort in engaging in research. This can be done by better informing society about the aims and processes of clinical research (Sacristán, 2016). The PHAC and Public Health Ontario can lead the collaboration among health care professionals, policymakers, social media, patient community organizations, and pharmaceutical companies to educate the society on information about the purposes of conducting research and the mechanisms available to protect participants, for example, the local REB (HiREB) in Hamilton (Sacristán, 2016). Enhancing the social image of research is needed (Sacristán, 2016). It should be made clear to the patients that new knowledge to benefit patients depends on the patients who participated in research conducted in the past (Sacristán, 2016). More effort is also needed in disseminating and applying research findings. When this is done well, it helps to improve the social stigmatization of autism and helps the community

to understand the importance for them to engage in research. Some of the KT strategies that can utilize to enhance the implementation of this intervention are by conducting campaigns in the media and organizing educational materials written in layman's language for the public. It is suggested that the stigmatizing cues and framing techniques used by journalists have contributed to the threat of autism in our society (Holton, 2014). Consequently, although the journalists are typically not considered as taking up an important role in clinical research, future collaborations with them are vital in enhancing patient engagement in research. Autism Ontario, the Canadian Autism Spectrum Disorder Alliance and Autism Speaks Canada, are some of the organizations in Canada that work towards raising public awareness of autism by recognizing the challenges that autistic individuals face in daily life. One of the key events that these organizations celebrate to break down autism stigma is the World Autism (Awareness) Day (April 2 of each year) or World Autism Month (April). For instance, Autism Ontario's theme for this year's event was "Celebrate the Spectrum" (Autism Ontario, n.d.). It engaged the community by organizing the "Design Your Flag Contest" (Autism Ontario, n.d.).

The Government of Canada invested $1.46 million for the PHAC to lead the development of a national autism strategy in collaboration with the Canadian Academy of Health Sciences, which is an evidence-based report on autism from autistic individuals, their families, and other relevant stakeholders (PHAC, 2020). As part of this project, a national autism conference will be conducted in fall 2022 (November 15 and 16, 2022) to solidify the priorities in the national autism strategy (PHAC, 2022). This is a good opportunity to emphasize the importance of engaging in research among the autistic communities and work towards removing the stigmatization of autism in Canadian society. At this time, the details of the national autism conference have not yet been released (PHAC, 2022). I would suggest the organizers to put in the effort of engaging different members of the community, rather than keeping the conference too scientific and professional as this will limit participation from the

autistic community. I would recommend asking the presenter to also include a paragraph of the research content in laymen's language. The organizers can increase the number of community members attending the conference by using part of the budget to provide financial incentives to attract them to attend. There can also be interactive sessions for community members and other relevant stakeholders to share their opinions and for educational purposes.

3.3 Final Remarks

Marginalized autistic populations, especially autistic children, are under-represented in research. The facilitators and barriers contributing to this unbalanced patient engagement in research from the perspective of community organizations, researchers and funders were assessed, and corresponding recommendations were made. More attention is needed to better engage autistic children with marginalized identities, especially the way how research sampling was done needs to be improved. Instead of engaging autistic children's parents/caregivers, engaging autistic children's first voice in research whenever possible is vital in revealing accurate information about them. Collaborations from policymakers, researchers and funders are needed to increase equity and enhance the applicability of research conducted in the autism field.

3.4 References

American Psychiatric Association. (2013). *DSM-V: Diagnostic and Statistical Manual of Mental Disorders*. http://archive.org/details/diagnosticstatis0005unse

Armstrong, R., Waters, E., Dobbins, M., Anderson, L., Moore, L., Petticrew, M., Clark, R., Pettman, T. L., Burns, C., Moodie, M., Conning, R., & Swinburn, B. (2013). Knowledge translation strategies to improve the use of evidence in public health decision making in local government: Intervention design and implementation plan. *Implementation Science*, *8*(1), 121. https://doi.org/10.1186/1748-5908-8-121

Autism Ontario. (n.d.). *World Autism Day*. Retrieved April 7, 2022, from https://www.autismontario.com/programs-services/world-autism-day

Beggiato, A., Peyre, H., Maruani, A., Scheid, I., Rastam, M., Amsellem, F., Gillberg, C. I., Leboyer, M., Bourgeron, T., Gillberg, C., & Delorme, R. (2017). Gender differences in autism spectrum disorders: Divergence among specific core symptoms. *Autism Research*, *10*(4), 680–689. https://doi.org/10.1002/aur.1715

Canadian Institutes of Health Research Government of Canada. (2005, October 27). *Knowledge translation—CIHR*. https://cihr-irsc.gc.ca/e/29529.html

Canadian Institutes of Health Research Government of Canada. (2016, September 8). *Health Research Training at CIHR - CIHR*. https://cihr-irsc.gc.ca/e/50507.html

CanChild. (2022). *Family Engagement in Research Course*. https://www.canchild.ca/en/research-in-practice/family-engagement-in-research-course

Crowe, S., Fenton, M., Hall, M., Cowan, K., & Chalmers, I. (2015). Patients', clinicians' and the research communities' priorities for treatment research: There is an important mismatch. *Research Involvement and Engagement, 1(1),* 2. *https://doi.org/10.1186/s40900-015-0003-x*

European Patients' Academy on Therapeutic Innovation. (n.d.). *EUPATI Open Classroom*. EUPATI. Retrieved April 8, 2022, from https://eupati.eu/training/medicines-rd/

Haney, J. L. (2016). Autism, females, and the DSM-5: Gender bias in autism diagnosis. *Social Work in Mental Health*, *14*(4), 396–407. https://doi.org/10.1080/15332985.2015.1031858

Holmes, B., Scarrow, G., & Schellenberg, M. (2012). Translating evidence into practice: The role of health research funders. *Implementation Science*, *7*(1), 39. https://doi.org/10.1186/1748-5908-7-39

Holton, A. E., Farrell, L. C., & Fudge, J. L. (2014). A Threatening Space?: Stigmatization and the Framing of Autism in the News. *Communication Studies*, *65*(2), 189–207. https://doi.org/10.1080/10510974.2013.855642

Interagency Advisory Panel on Research Ethics Government of Canada. (2019, April 1). *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans – TCPS 2 (2018) – Chapter 4: Fairness and Equity in Research Participation*. https://ethics.gc.ca/eng/tcps2-eptc2_2018_chapter4-chapitre4.html#6

Interagency Advisory Panel on Research Ethics Government of Canada. (2019, April 1). *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans – TCPS 2 (2018) – Chapter 7: Conflicts of Interest*. https://ethics.gc.ca/eng/tcps2-eptc2_2018_chapter7-chapitre7.html

Interagency Advisory Panel on Research Ethics Government of Canada. (2019, April 1). *Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans – TCPS 2 (2018) – Chapter 9: Research Involving the First Nations, Inuit and Métis Peoples of Canada*. https://ethics.gc.ca/eng/tcps2-eptc2_2018_chapter9-chapitre9.html

Khan, A. Y., Preskorn, S. H., & Baker, B. (2005). Effect of Study Criteria on Recruitment and Generalizability of the Results. *Journal of Clinical Psychopharmacology*, *25*(3), 271–275. https://doi.org/10.1097/01.jcp.0000161497.73514.80

Pitts, M. J., & Miller-Day, M. (2007). Upward turning points and positive rapport-development across time in researcher—Participant relationships. *Qualitative Research*, *7*(2), 177–201. https://doi.org/10.1177/1468794107071409

Pohl, A. L., Crockford, S. K., Blakemore, M., Allison, C., & Baron-Cohen, S. (2020). A comparative study of autistic and non-autistic women's experience of motherhood. *Molecular Autism*, *11*(1), 3. https://doi.org/10.1186/s13229-019-0304-2

Public Health Agency of Canada. (2016, January 18). *Signs and symptoms of autism spectrum disorder (ASD)* [Education and awareness]. https://www.canada.ca/en/public-health/services/diseases/autism-spectrum-disorder-asd/signs-symptoms-autism-spectrum-disorder-asd.html

Public Health Agency of Canada. (2020, October 28). *Autism: National autism strategy* [Education and awareness]. https://www.canada.ca/en/public-health/services/diseases/autism-spectrum-disorder-asd/national-strategy.html

Public Health Agency of Canada. (2022, April 1). *National Autism Conference* [Promotional material]. https://www.canada.ca/en/public-health/services/diseases/autism-spectrum-disorder-asd/national-strategy/conference-2022.html

Sacristán, J. A., Aguarón, A., Avendaño-Solá, C., Garrido, P., Carrión, J., Gutiérrez, A., Kroes, R., & Flores, A. (2016). Patient involvement in clinical research: Why, when, and how. *Patient Preference and Adherence*, *10*, 631–640. https://doi.org/10.2147/PPA.S104259

Statistics Canada. (2017). 2016 Census. Statistics Canada Catalogue no. 98-316-X2016001. Ottawa. *Hamilton, C [Census subdivision], Ontario and Hamilton, CDR [Census*

division], Ontario (table). *Census Profile*. Retrieved April 8, 2022, from https://www12.statcan.gc.ca/census-recensement/2016/dp-pd/prof/index.cfm?Lang=E

Werling, D. M., & Geschwind, D. H. (2013). Sex differences in autism spectrum disorders. *Current Opinion in Neurology*, *26*(2), 146–153. https://doi.org/10.1097/WCO.0b013e32835ee548

Wilkins, C. H. (2018). Effective Engagement Requires Trust and Being Trustworthy. *Medical Care*, *56*(10 Suppl 1), S6. https://doi.org/10.1097/MLR.0000000000000953