Linda Nguyen*, Susan M. Jack, Briano Di Rezze, Marjolijn Ketelaar and Jan Willem Gorter

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

<https://doi.org/10.1515/jtm-2021-0004>

Received April 16, 2021; accepted August 8, 2021;

published online September 6, 2021

Abstract

Background: Children and youth with neurodisabilities may experience different challenges during their transition to adulthood, such as pursuing postsecondary education, finding employment, and navigating a new adult health care system. Families, including siblings, have an important role in the process for when youth with neurodisabilities are transitioning to adulthood. Siblings are in a unique position, where they can have different roles such as a friend, mentor, or caregiver. Siblings can offer various supports to their brother or sister with a neurodisability, but

they require knowledge and skills for these different supporting roles. Currently, there are limited programs available for siblings to learn how to support their brother or sister with a neurodisability during transition. A first step to develop these sibling support programs is to understand the experiences of siblings of youth with a neurodisability. The purpose of this report is to describe a protocol of a qualitative case study aimed at examining the experiences of siblings including their roles and responsibilities to their brother or sister with a neurodisability during health care transition.

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

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

Keywords: disability; health care transition; qualitative study; sibling.

***Corresponding author: Linda Nguyen**, PhD Candidate, School of Rehabilitation Science, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; and CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada, E-mail: nguyel7@mcmaster.ca. <https://orcid.org/0000-0002-7165-6678>

Susan M. Jack, School of Nursing, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; Department of Health Research Methods, Evidence, and Impact, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; and Offord Centre for Child Studies, Hamilton, Ontario, Canada. <https://orcid.org/0000-0003-4380-620X>

Briano Di Rezze, School of Rehabilitation Science, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; and CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada. <https://orcid.org/0000-0002-6061-2506>

Marjolijn Ketelaar, CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada; and Centre of Excellence for Rehabilitation Medicine, University Medical Center Utrecht and De Hoogstraat Rehabilitation, Utrecht, Netherlands. <https://orcid.org/0000-0002-8324-518X>

Jan Willem Gorter, School of Rehabilitation Science, Faculty of Health Sciences, McMaster University, Hamilton, Ontario, Canada; CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada; and Department of Pediatrics, McMaster University and McMaster Children's Hospital, Hamilton, Ontario, Canada. <https://orcid.org/0000-0002-3012-2119>

Introduction

Youth have numerous opportunities to explore their future interests and goals, including school, work, family, and

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

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

potential unique roles that siblings can have during health care transition.

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

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

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

possible gaps and topics to address in the development of future resources or tools to support youth and young adult siblings of individuals with a neurodisability.

Objectives

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

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

Integrated knowledge translation

Integrated knowledge translation is an approach to doing research with knowledge users, such as patients and families, as equal partners with researchers throughout the study [19]. Siblings with the lived experiences of having a brother or sister with a disability have often described the

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

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

Case study methodology

A descriptive single case study design will be used to understand the roles and responsibilities of siblings to support their brother or sister with a neurodisability in the preparation for health care transition [23]. This qualitative case study design is an empirical approach that

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

Designing the case

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

Sibling relationships

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

Childhood-onset neurodisabilities

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

Health care transition

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

Case binding

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

Geographical location

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

Timeframe

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

Conceptual framework

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

- (1) *Transition theory* [33]: Meleis and colleagues developed the transition theory to describe the different types of transition, the properties of the transition experience, and how individuals experience the process of transition resulting in specific outcomes [33]. Specific properties or characteristics of the sibling relationship, such as the emotions and attitudes of siblings towards each other as well as the level of closeness and frequency of contact between siblings [9, 34], can influence how siblings react to the trigger of health care transition and the outcomes that occurs regarding their roles and responsibilities.
- (2) *Bioecological systems theory* [35]: Urie Bronfenbrenner, a developmental psychologist, developed this theory to describe person-environment transactions, where the interaction between the youth with a neurodisability and multiple environments is a transactional process. This study will focus on the microsystem that encompasses individuals within the immediate environment of the TD sibling including parents, siblings, and health care providers as well as the mesosystem that includes the interactions between the microsystems such as the relationships with TD siblings, family members, and health care providers. Family roles could change when a youth with a neurodisability is preparing for health care transition [36]. There could also be changes in the sibling relationship, which is influenced by the context of the family that exists within the broader system of the external systems of health care, school, community, and policies. Figure 1 presents the initial conceptual framework, which will continue to evolve as the data is collected and analyzed. The final framework will include findings that address the propositions and constructs.

Propositions

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

- (1) *Gender*. Sibling dyads of the same gender are more likely to model one another, because they view similarities with each other [38]. We recognize that gender is non-binary and that siblings may identify themselves along a spectrum. In our study, we refer to siblings as a “brother” or “sister” with the recognition that we will ask study participants about how they prefer to identify themselves.
- (2) *Age*. Siblings who are older than their brother or sister with a neurodisability are likely to take on caregiving roles compared to younger siblings [7].
- (3) *Health*. There may be differences in sibling roles depending on the overall health status and neurodisability diagnosis of their brother or sister [39].

Participant recruitment and sampling strategies

Participants will be recruited based on the following inclusion criteria: 1) between 14 and 40 years old; 2) able to speak English; and 3) has a brother or sister with a diagnosis of a childhood-onset neurodisability, such as autism spectrum disorder, cerebral palsy, Down syndrome, epilepsy, fetal alcohol spectrum disorder, or spina bifida between 14 and 21 years old. While adult siblings will be recruited in this study, they will be asked to reflect on their experiences during adolescence and how their roles and responsibilities to their brother or sister with a neurodisability might have changed over time. Participants will be recruited using social media, in which a website [40] has been created to share the study information. The recruitment materials include a study poster [41] and video that was co-created with the SibYAC [42]. These materials will be posted with a recruitment message on different social media platforms, such as Facebook, Twitter, and Instagram, and shared using the networks from the research team and reaching out to disability-specific organizations. A summary of how the recruitment materials have been shared is presented in Table 1. This study will also reach out to potential sibling participants

Figure 1: Initial conceptual framework.

of the brother or sister with a neurodisability from Ontario who is participating in an ongoing multi-regional Canadian randomized controlled trial [43]. In addition, snowball sampling will be used, in which participants in the study can refer other siblings [44]. Convenience and theoretical sampling will be conducted while data collection and ongoing data analysis is being conducted. Theoretical sampling allows for additional participants to be purposefully recruited who can provide in-depth rich information about the emerging codes, categories, and themes that may arise during data analysis [45]. Co-investigators, SibYAC members, and sibling participants will be asked to recruit potential siblings through their social networks. This study was approved by the Hamilton Integrated Research Ethics Board (REB Project 7932).

Sample size

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

Data collection

In this study, data will be collected through semi-structured, 1:1 in-depth interviews augmented by photo elicitation. Our SibYAC identified the novelty of photo elicitation to increase engagement with siblings as participants. They also believed that photo elicitation would enhance the dissemination of study findings with the potential to share photos through other avenues, such as a video to be posted on social media. Each participant will be invited to participate in a single interview of approximately 60–90 min. Due to public health measures and physical distancing guidelines from COVID-19 [49], all interviews will be conducted by phone or by videoconferencing (Zoom Communications Inc.) and participants may choose to turn on their web camera. During the interview, photo elicitation techniques will be used to deepen the nature of the discussion about their experiences with their sibling. Photo elicitation can be used with young adults to enhance their level of engagement in studies, especially since they are familiar with photography through their use of technology on social media, such as Instagram [50]. Photographs can help raise topics that may not necessarily be verbally raised [51]. This study will use photo elicitation by asking participants to bring approximately 3–5 pre-existing photographs in preparation for the interview that exemplify their sibling experiences,

Table 1: A description of recruitment strategies.

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

including their roles and responsibilities related to supporting their brother or sister’s preparation for health care transition. Electronic consent will be obtained from participants through Research Electronic Data Capture (REDCap™) [52], which is a secure web application to create and manage online forms (www.project-redcap.org) and will be hosted by the Department of Pediatrics at McMaster University. The consent process includes the following three steps [53]: i) consent to participate in the interview, including a description about their participation, benefits, and risks; ii) permission if participants share photographs from their parents or sibling; and iii) permission to share photographs on selected platforms (e.g., study publication, scientific presentations at conferences, workshops, videos, lay summaries, infographics). Steps two and three are optional, in which participants may choose to share photographs during the interview if they would like, but they can continue to participate in a verbal interview without sharing photographs. Participants can share the photographs before the interview by uploading them onto a

secure cloud storage provided through McMaster’s Mac-Drop (<https://drop.mcmaster.ca/login>). For all participants, the transcripts will be deanonymized to ensure confidentiality, and only the verbal descriptions of the photographs will be included in the analysis of the transcripts.

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

Data management

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

Data analysis

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

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

Discussion

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

A second strength of this study is the use of social media as a recruitment strategy in order to adapt to physical distancing guidelines resulting from the COVID-19 pandemic. This study will document the different social

media platforms that have been successful in reaching the audience of youth and young adult siblings who participated in this study. These recruitment strategies can inform future research even when physical distancing restrictions are relaxed.

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

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

Conclusion

Siblings share a lifelong bond with their brother or sister, and siblings have a unique relationship in which they grow up together in the same home environment. When a

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

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

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

Competing interests: Authors state no conflict of interest.

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

Informed consent: Not applicable.

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

References

1. Stewart D. Transition to adult services for young people with disabilities: current evidence to guide future research. *Dev Med Child Neurol* 2009;51(4 Suppl):169–73.
2. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. *Lancet Child Adolesc Health* 2018;2:223–8.
3. Freeman M, Stewart D, Cunningham CE, Gorter JW. Information needs of young people with cerebral palsy and their families during the transition to adulthood: a scoping review. *J Transit Med* 2018;1:20180003.
4. Blum RW. Improving transition for adolescents with special health care needs from pediatric to adult-centered health care: introduction. *Pediatrics* 2002;110:1301–3.
5. Stewart DA, Law MC, Rosenbaum P, Willms DG, Stewart DA, Law MC, et al. A qualitative study of the transition to adulthood for youth with physical disabilities. *Phys Occup Ther Pediatr* 2009;21:3–21.
6. Whiteman SD, McHale SM, Soli A. Theoretical perspectives on sibling relationships. *J Fam Theory Rev* 2011;3:124–39.
7. Hall SA, Rossetti Z. The roles of adult siblings in the lives of people with severe intellectual and developmental disabilities. *J Appl Res Intellect Disabil* 2018;31:423–34.
8. Corsano P, Musetti A, Guidotti L, Capelli F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *J Intellect Dev Disabil* 2017;42:151–61.
9. Atkin K, Tozer R. Personalisation, family relationships and autism: conceptualising the role of adult siblings. *J Soc Work* 2014;14:225–42.
10. The Sibling Collaborative. Understanding the sibling experience: sibling needs assessment survey 2017 [Internet]; 2018. 1–19. Available from: [http://cdn.agilitycms.com/partners-for-planning/Understanding the Sibling Experience–March 2018–Final.pdf](http://cdn.agilitycms.com/partners-for-planning/Understanding%20the%20Sibling%20Experience-March%202018-Final.pdf) [Accessed 2 Jun 2021].
11. Moyson T, Roeyers H. "The overall quality of my life as a sibling is all right, but of course, it could always be better". Quality of life of siblings of children with intellectual disability: the siblings' perspectives. *J Intellect Disabil Res* 2012;56:87–101.
12. Arnold CK, Heller T, Kramer J. Support needs of siblings of people with developmental disabilities. *Intellect Dev Disabil* 2012;50:373–82.
13. McKenzie Smith M, Pinto Pereira S, Chan L, Rose C, Shafran R. Impact of well-being interventions for siblings of children and young people with a chronic physical or mental health condition: a systematic review and meta-analysis. *Clin Child Fam Psychol Rev* 2018;21:246–65.
14. Sibling Support Project. The sibling support project [Internet]; 2020. Available from: <https://www.siblingsupport.org/> [Accessed 15 Nov 2020].
15. Opening Hearts. SibTeens [Internet]; 2019. Available from: <https://www.openinghearts.ca/sibteens/> [Accessed 2 Jun 2021].
16. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M, et al. Developing and evaluating complex interventions: the new Medical Research Council guidance. *BMJ* 2008;337:a1655.
17. Corsano P, Musetti A, Guidotti L, Capelli F. Typically developing adolescents' experience of growing up with a brother with an autism spectrum disorder. *J Intellect Dev Disabil* 2017;42:151–61.
18. Nguyen T, Gorter JW. Use of the international classification of functioning, disability and health as a framework for transition from paediatric to adult healthcare. *Child Care Health Dev* 2014;40:759–61.
19. Canadian Institutes of Health Research. Guide to knowledge translation planning at CIHR: integrated and end-of-grant approaches [Internet]; 2015. Available from: <http://www.cihr-irsc.gc.ca/e/45321.html#a3> [Accessed 22 Apr 2019].
20. Richardson SLL, Jordan LS. Qualitative inquiry of sibling relationships: reinforcement of disability devaluation through the exclusion of voices. *Disabil Soc* 2017;32:1534–54.
21. Smits D-W, van Meeteren K, Klem M, Alsem M, Ketelaar M. Designing a tool to support patient and public involvement in research projects: the involvement Matrix. *Res Involv Engagem* 2020;6:30.
22. CHILD-BRIGHT Network. Guidelines for patient partner compensation; 2020. Available from: <https://www.child-bright.ca/compensation-guidelines> [Accessed 23 Jun 2021].
23. Yin RK. Designing case studies. In: Case study research and applications: design and methods, 6th ed. Thousand Oaks, California: SAGE; 2018:25–80 pp.

24. Yin RK. Getting started. In: Case study research and applications: design and methods, 6th ed. Thousand Oaks, California: SAGE; 2018:3–24 pp.
25. Kingsnorth S, Rudzik AEF, King G, McPherson AC. Residential immersive life skills programs for youth with disabilities: a case study of youth developmental trajectories of personal growth and caregiver perspectives. *BMC Pediatr* 2019;19:413.
26. Turcotte P-L, Côté C, Coulombe K, Richard M, Larivière N, Couture M. Social participation during transition to adult life among young adults with high-functioning autism spectrum disorders: experiences from an exploratory multiple case study. *Occup Ther Ment Health* 2015;31:234–52.
27. American Academy of Pediatrics. Sibling relationships [Internet]; 2007. Available from: https://www.acy.ac.cy/nursery/documents/Sibling_Relation.pdf [Accessed 19 Apr 2019].
28. Morris C, Janssens A, Tomlinson R, Williams J, Logan S. Towards a definition of neurodisability: a Delphi survey. *Dev Med Child Neurol* 2013;55:1103–8.
29. CHILD-BRIGHT Network. READYorNot(TM) brain-based disabilities project [Internet]; 2019. Available from: <https://www.child-bright.ca> [Accessed 15 Nov 2020].
30. Canadian Paediatric Society. Positon statement (AH 2007-01). Transition to adult care for youth with special health care needs. *Paediatr Child Health* 2007;12:785–8.
31. Baxter P, Jack S. Qualitative case study methodology: study design and implementation for novice researchers. *Qual Rep* 2008;13:544–59.
32. Gorter JW, Stewart D, Smith MW, King G, Wright M, Nguyen T, et al. Pathways toward positive psychosocial outcomes and mental health for youth with disabilities: a knowledge synthesis of developmental trajectories. *Can J Community Ment Health* 2014; 33:45–61.
33. Meleis AI, Sawyer LM, Im EO, Messias DK, Schumacher K. Experiencing transitions: an emerging middle-range theory. *Adv Nurs Sci* 2000;23:12–28.
34. Davys D, Mitchell D, Haigh C. Futures planning, parental expectations and sibling concern for people who have a learning disability. *J Intellect Disabil* 2010;14:167–83.
35. Bronfenbrenner U. Six theories of child development: revised formulations and current issues. In: Vasta R, editor. *Ecological systems theory*. London: Jessica Kingsley Publishers; 1992: 187–249 pp.
36. Palisano RJ, Di Rezze B, Stewart D, Rosenbaum PL, Hlyva O, Freeman M, et al. Life course health development of individuals with neurodevelopmental conditions. *Dev Med Child Neurol* 2017;59:470–6.
37. Yin RK. Designing case studies: identifying your case(s) and establishing the logic of your case study. In: Case study research and applications: design and methods, 6th ed. Thousand Oaks, California: Sage Publications Inc.; 2018:25–78 pp.
38. Bandura A. Toward a unifying theory of behavioral change. *Psychol Rev* 1977;84:191–215.
39. Orsmond GI, Kuo H-Y, Seltzer MM. Siblings of individuals with an autism spectrum disorder. *Autism* 2009;13:59–80.
40. CanChild Centre for Childhood Disability Research. BrothErs and Sisters involvement in health care TranSition for youth with Brain-based disabilities (BEST SIBS) Study [Internet]; 2021. Available from: <https://canchild.ca/en/research-in-practice/current-studies/brothers-and-sisters-involvement-in-health-care-transition-for-youth-with-brain-based-disabilities-best-sibs-study> [Accessed 11 Apr 2021].
41. CanChild Centre for Childhood Disability Research. BEST SIBS study poster; 2021. Available from: [https://canchild.ca/system/tenon/assets/attachments/000/003/528/original/Recruitment_Poster_\(clean_copy\)_-_February_8__2021.pdf](https://canchild.ca/system/tenon/assets/attachments/000/003/528/original/Recruitment_Poster_(clean_copy)_-_February_8__2021.pdf) [Accessed 11 Apr 2021].
42. CanChild Centre for Childhood Disability Research. BEST SIBS study recruitment video; 2021. Available from: <https://vimeo.com/522169935/1e2b08984d> [Accessed 11 Apr 2021].
43. Gorter JW, Amaria K, Kovacs A, Rozenblum R, Thabane L, Galuppi B, et al. CHILD-BRIGHT READYorNot Brain-Based Disabilities Trial: protocol of a randomised controlled trial (RCT) investigating the effectiveness of a patient-facing e-health intervention designed to enhance healthcare transition readiness in youth. *BMJ Open* 2021;11:48756.
44. Patton MQ. Designing qualitative studies. In: *Qualitative evaluation and research methods*, 2nd ed. Newbury Park: Sage Publications Inc.; 1990:145–98 pp.
45. Robinson OC. Sampling in interview-based qualitative research: a theoretical and practical guide. *Qual Res Psychol* 2014;11: 25–41.
46. Nguyen L, Davis H, Bellefeuille S, Ketelaar M, Di Rezze B, Jack S, Gorter JW, Sibling Youth Advisory Council. Sibling engagement in research: An illustrative example of a sibling-researcher partnership to design a study. CHILD-BRIGHT virtual symposium part 2. [Internet]; 2020. Available from: <https://www.child-bright.ca/events/2020/4/29/symposium-pt2> [Accessed 11 Apr 2021].
47. Nguyen L, Ketelaar M, Di Rezze B, Jack S, Gorter JW, Sibling Youth Advisory Council. The BEST SIBS study: a qualitative case study to understand the experiences of siblings of youth with a brain-based disability during health care transition [Internet]; 2021. Available from: <https://www.childrenshealthcarecanada.ca/transitions-poster-hall/2021/1/11/the-best-sibs-study-a-qualitative-case-study-to-understand-the-experiences-of-siblings-of-youth-with-a-brain-based-disability-during-health-care-transition> [Accessed 11 Apr 2021].
48. Avieli H, Band-Winterstein T, Araten Bergman T. Sibling relationships over the life course: growing up with a disability. *Qual Health Res* 2019;29:1739–50.
49. Government of Canada. Coronavirus disease (COVID-19) [Internet]; 2020. Available from: <https://www.canada.ca/en/public-health/services/diseases/coronavirus-disease-covid-19.html> [Accessed 23 Jun 2021].
50. Sibeoni J, Costa-Drolon E, Poulmarc'h L, Colin S, Valentin M, Pradère J, et al. Photo-elicitation with adolescents in qualitative research: an example of its use in exploring family interactions in adolescent psychiatry. *Child Adolesc Psychiatr Ment Health* 2017; 11:1–11.
51. Croghan R, Griffin C, Hunter J, Phoenix A. Young people's constructions of self: notes on the use and analysis of the photo-elicitation methods. *Int J Soc Res Methodol* 2008;11: 345–56.
52. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)—a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inf* 2009; 42:377–81.

53. Wang CC, Redwood-Jones YA. Photovoice ethics: perspectives from flint photovoice. *Health Educ Behav* 2001;28:560–72.
54. Bagnoli A. Beyond the standard interview: the use of graphic elicitation and arts-based methods. *Qual Res* 2009;9:547–70.
55. Kearney KS, Hyle AE. Drawing out emotions: the use of participant-produced drawings in qualitative inquiry. *Qual Res* 2004;4:361–82.
56. Birks M, Francis K. Memoing in qualitative research probing data and processes. *J Res Nurs* 2008;13:68–75.
57. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.
58. Redquest BK, Tint A, Ries H, Lunskey Y. Exploring the experiences of siblings of adults with intellectual/developmental disabilities during the COVID-19 pandemic. *J Intellect Disabil Res* 2021;65: 1–10.
59. Statistics Canada. Population estimates, quarterly [Internet]; 2021. Available from: <https://www150.statcan.gc.ca/t1/tbl1/en/tv.action?pid=1710000901> [Accessed 11 Apr 2021].