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Research Involvement
and Engagement

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The co-development and pilot evaluation of the Siblings Training, Empowerment, and Advocacy Kit (Siblings TEAKit) to support youth and young adult siblings of individuals with a disability: A participatory action research qualitative study protocol

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Abstract

Introduction Siblings can have multiple roles in the lives of each other over time, including the roles of friend, role model, and caregiver at different time points. These roles may be different for siblings of youth with disabilities, and yet, there are limited resources to support siblings in the health, rehabilitation, and education systems. This protocol outlines a study for which the primary aim is to develop and evaluate a toolkit to support youth and young adult siblings to initiate conversations about their roles and responsibilities with their sibling(s) with a disability and family. The secondary aim of the study is to test a framework for and evaluate the process of partnered research with siblings and other family members.

Methods This qualitative descriptive study will adopt a participatory approach where a Sibling Youth Advisory Council (SibYAC) of young adult siblings of individuals with a disability and/or chronic health condition will be engaged as research partners. The long standing partnership with the SibYAC and our team has informed the need to conduct this study. This study will be conducted in two stages to address our primary aim. First, a co-design workshop will be held with siblings (ages 14 to 25 years old) to develop the content and format of the toolkit. Second, focus groups will be conducted with siblings, parents, and healthcare professionals to provide feedback on the toolkit prototypes. Data will be analyzed using directed content analysis with implementation science frameworks as a guide. Specifically, the Theoretical Domains Framework and COM-B model will be used to identify toolkit elements focused on supporting siblings, and the Consolidated Framework for Implementation Research (CFIR) will provide considerations about settings for where the toolkit could be implemented in the future. The secondary aim is addressed where our partnership with the SibYAC will be evaluated through formative discussions and the Public and Patient Engagement Evaluation Tool.

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Discussion This study protocol advances the methods and rigour in participatory research with young people, including siblings. This study is the first step towards the development of a toolkit to support siblings of persons with disabilities to foster dialogue between families, health and education professionals about their roles. Clear expectations about family roles can promote positive health and wellbeing for siblings and the whole family.

Plain English summary

Siblings can have different roles in the lives of each other, such as a friend, role model, or caregiver. There are few supports for siblings who are youth and young adults of individuals with disabilities in the fields of health, rehabilitation, and education. This manuscript describes the process to creating and testing a toolkit to help youth and young adult siblings with their sibling(s) with a disability and family. In this study, we partnered with siblings as members of our research team who highlighted the importance of conducting this study. This study will be conducted in two parts. First, a workshop will be held with siblings (ages 14 to 25 years old) to discuss information to be included in the toolkit and how the toolkit should look like. Second, we will have conversations with groups of siblings, parents, and healthcare professionals who can provide feedback on the toolkit. We will analyze the data to see what kind of information should be included in the toolkit and how the toolkit can be delivered. We will use tools to study how we can improve our partnership with siblings and researcher. This study is the first step towards creating a toolkit to support siblings of persons with disabilities. The toolkit can guide siblings in having conversations with their families and healthcare professionals about their roles. It is important to have open and clear communication about the roles that siblings and families have, ultimately supporting positive health and wellbeing of siblings and the whole family.

Background

In North America, approximately 15% to 18% of youth (ages 0 to 17 years old) have a disability and/or chronic health condition [1, 2]. During the transition from adolescence to adulthood, defined as the period from ages 18 to 25 years old [3], youth with a disability will learn from multiple experiences, including learning how to form relationships with peers and partners, graduating from high school, entering post-secondary education and/or the workforce, and navigating adult care services. Youth will also learn how to become more independent in activities and roles in life [4]. During this critical period of 'emerging adulthood' youth can turn to their families, including siblings, for different types and levels of support as they navigate the transition to emerging adulthood.

Siblings have a unique relationship. They usually grow up in the same home environment and are fellow travelers in each other's life journey [5, 6]. When raised in the same household, siblings may have a unique understanding of habits and preferences, health care history and concerns pertinent to one another [7]. Siblings have described the challenges that they may experience growing up with their sibling with a disability, such as having less parental attention and learning how to be independent from a young age [8–10]. Siblings may also feel guilty when they are not available to support the family, such as if they have other social plans, or if they have to move away from home for employment or higher education [11, 12]. Yet, siblings also share the positive experiences from their sibling, such as the strong connectedness with the family, learnings that they have from their sibling, developing empathy and awareness about disability, and deciding on careers based on their personal experiences [13–15].

The relationship that siblings have with each other can change over time [16, 17]. When a sibling has a disability, siblings often recognize from a young age that they may have a support role for their sibling with a disability and their family [9, 11]. For example, they may provide peer support in school, be present at their sibling's medical appointments, or support their parents and/or caregivers with care tasks in the home when required [9, 11, 18]. There are numerous roles that siblings take on and factors that impact the development of their roles over time [19]. In an earlier study we conducted, siblings identified specific roles that they have with their sibling with a disability such as: being a friend, role model/mentor, protector, advocate, supporter, and caregiver [16, 17]. Sibling participants identified that certain characteristics influenced the types of roles that they chose, including their personal identity, values and experiences. For example, some sibling participants disclosed that they had a disability themselves, and they were motivated to support their sibling with a disability because they had to overcome challenges that their sibling with a disability was facing. The research literature on siblings supports these findings, and also described other factors that can influence the roles that siblings choose to have [20, 21]. The personal characteristics of the siblings with and without a disability (e.g., the temperament of a sibling can affect how they managed their conflicts and stresses within a relationship [22–24]), the health condition of the sibling (e.g., the level of care required [25, 26]), and gender of the siblings (e.g., females have been perceived to be more involved in the sibling relationship) [27, 28] are some factors that influence this relationship. Family factors could also influence the nature of the sibling relationship, such as the health of the parents and/or caregivers, experiences of caregiver burden, and socioeconomic status [29, 30]. Having consistent and regular family routines and problem solving skills in the family can support siblings [31].

As siblings enter adolescence and emerging adulthood, their relationship with their sibling with a disability might change. Siblings might become closer with each other where some siblings begin to develop an understanding about the disability and may frequently connect with each other throughout the life course [5, 20, 32]. Some siblings might naturally understand each other and identify as being lifelong companions [9, 33]. Depending on the age gap, siblings may be navigating their own developmental trajectory. For example, siblings might have to move away from home for employment, post-secondary education, or personal reasons [5]. Siblings might also start to build a life outside of their immediate family with new experiences and personal relationships, expanding the relationship circle of the family, but also limiting the availability and time spent within the nuclear family [5]. In previous studies, we identified that sibling participants as young as 14 years have indicated that while they did not have conversations about their roles with their family, they anticipated having an active role such as being a caregiver to their sibling with a disability in the future [16]. Findings from other studies indicated that siblings wanted to have more discussions with their family about their roles [8, 34]. There is a need for families to have open discussions and clear expectations, which might help siblings to feel prepared for their future roles, as well as increase communication about the perspectives from both parents and siblings about their roles [35]. While families may have discussions to plan for the future, these plans often do not define the activities that siblings might be expected to take on [36].

While siblings dynamics are a natural part of family roles and expectations, there is limited information about how families and healthcare providers can support siblings in the preparation for the roles they might naturally or be expected have. A recent review and qualitative document analysis of resources available for siblings in Canadian children's hospitals, rehabilitation centres, and treatment centres identified the lack of resources to support siblings in having conversations with their family about their roles [19]. This review identified that there are few resources for siblings to refer to when learning about how to be involved with the healthcare of their sibling with a disability. An example of these resources include a toolkit [37] for siblings to learn about the diagnosis of the disability of their sibling. Young siblings (ages 8 to 13 years old) can attend Sibshops to connect with other siblings who have a sibling with a disability [38]. A scoping review of programs to support siblings identified programs that included sessions for the whole family to attend [39]. For example, a program tailored for siblings ages to 8 to 13 years old had them record a video about their experiences that parents reviewed afterwards [40].

Our team partnership and work to date

There is a need to understand the lived and living experiences of the study population to ensure that the research reflects their priorities and values. Throughout the process of designing and conducting this study, our team has partnered, and will continue to partner, with a Sibling Youth Advisory Council (SibYAC). The SibYAC was established in 2018 by the first author (LN) to partner in her doctoral dissertation. Members of the SibYAC are young adult siblings of youth with a disability and/or chronic health condition living in Canada. In our work to date, we have taken a non-categorical approach to view disability and/or chronic health conditions that shifts the focus from diagnoses to understanding the experiences, and the supports and services required for individuals [41–43]. The focus of our studies has included the experiences of siblings who have a sibling with a neurodevelopmental disability such as cerebral palsy and autism spectrum disorder [41, 44].

The SibYAC members were identified through word-of-mouth with researchers and national research networks, for example, the CanChild Centre for Childhood Disability Research [45], CHILD-BRIGHT Network [46], and Family Engagement in Research course [47]. The SibYAC have been meeting regularly through monthly meetings for the last six years over Zoom. The meetings are facilitated by the first author (LN) in this study, and the meeting agenda and topics of discussion were decided in collaboration by all members of the SibYAC.

We have been conducting our studies in partnership with the SibYAC within the context of a patient-oriented research network, where members of the team has been extensively involved with the CHILD-BRIGHT Network, a network funded under the Strategy for Patient-Oriented Research from the Canadian Institutes of Health Research [46]. Our partnership experiences includes working collaboratively in participatory action research studies centred on understanding the experiences of siblings who have a sibling with a disability and synthesizing resources to support them [16, 17, 19, 39, 48]. The application of a participatory action research approach provides guidance for how we engaged with the SibYAC to ensure that the purpose of this study, as well as the design and execution of the study, and the dissemination of the study findings, are guided by and relevant to the community of interested parties, specifically siblings and families [49–54]. Our work to date has been shared on our study website [55]. The involvement of the SibYAC throughout our previous work leading up to this study illustrates their empowerment towards conducting research that can foster impactful change among siblings, which is a key component of participatory action research [56]. Alongside our research studies, the SibYAC reflected on their experiences in requiring resources to support them in their roles and begin to have explicit conversations about their roles with their family. Based on these gaps, we designed this study to co-develop a toolkit to support siblings and provide a guide for beginning conversations in the family about their roles.

Based on research and lived and living experiences, the primary aim of this proposed study is to develop and evaluate a toolkit to support youth and young adult siblings to initiate conversations about their roles and responsibilities with their sibling(s) with a disability and family. The secondary aim of this study aims to evaluate the process of how we engaged in a partnership between researchers and siblings with lived and living experience. In this paper, we report on how we have partnered together using the Guidance for Reporting Involvement of Patients and the Public (GRIPP2) short form [57].

Methodology

Human-centered co-design

A human-centred co-design approach will be used to develop the toolkit. Human-centred co-design is an approach to understand how individuals interact with a product, system and/or service [58]. This approach is characterized by a clear and explicit understanding of the users, tasks, and environments, consideration of the user experience, and involvement of users throughout the design and development phases in an iterative process [58]. This approach can help to co-design a toolkit, Siblings Training, Empowerment, and Advocacy Kit (Siblings TEAKit), to support siblings based on their perspectives.

The phases of human-centred co-design [59] include the following:

1. Empathize: To understand the needs of siblings. Our team synthesized resources and completed the analysis of a qualitative study to understand the experiences of siblings of individuals with a disability [16, 19]. Through this phase we learned that siblings have limited conversations with their family to prepare for their roles.
2. Define: To identify a clear problem to be addressed. Based on the current literature and our previous work [16, 19], and in partnership with the siblings-partners we defined that a practical toolkit to facilitate conversation between parents, healthcare providers and siblings is the most needed problem to be addressed.
3. Ideate: To generate ideas about how to create innovative solutions. The current study protocol proposes to generate ideas for a toolkit with and for siblings.
4. Prototype: To generate different products as potential solutions to the problem. A prototype can be anything that a user can interact with, such as sticky notes, storyboard, or activity. Participants in this study will be asked to generate and provide feedback on prototypes for the toolkit.
5. Test: To evaluate the prototypes within the real-life context of the user. The current protocol proposes a pilot testing of the toolkit with the study participants and siblings-partners to assess feasibility and usability. A future study will conduct a thorough evaluation of the toolkit in different settings.

Development of toolkit

The Siblings Training, Empowerment, and Advocacy Kit (Siblings TEAKit) will be developed as an intervention tool to support siblings to initiate conversations with their family about planning and preparing for their roles. The development of the Siblings TEAKit will be conducted in two stages.

Stage 1. Co-design workshop

The purpose of the workshop is to define the content to be included in the Siblings TEAKit. The workshop will take approximately 4 h by Zoom video-conferencing software, and will be audio-recorded and transcribed. The content presented to participants will be based on research evidence on the important aspects and roles of siblings of youth with disabilities in the health care, education, and formal and informal supports offered to their sibling. The co-design workshop participants will be invited to reflect on the evidence-based content, and identify the most salient points, the areas that must be covered in a toolkit, and how the toolkit can be used to initiate conversation on the topics identified in previous research. Participants for the workshop will be siblings of youth with developmental disabilities who will be the users of the Siblings TEAKit. The activities for the workshop will be informed by a process guide [59] and field guide [60]. The process guide [59] outlines the overall process of the Ideate and Prototype phases in this study, and the field guide [60] further complements these phases with examples of activities to be completed.

Some examples of questions that we will ask in our workshop activities include:

1. If you could develop a toolkit as a resource for siblings that focused on how they could have a conversation with their family and healthcare professionals, what kind of topics would you include?
2. What might be helpful for the sibling to learn from these conversations?
3. What supports can be offered to siblings to prepare them for their roles to their sibling with a disability and family?

Stage 2. Focus groups

Following the co-design workshops, the purpose of the focus groups is to gain additional insights and feedback on the prototypes of the Siblings TEAKit. To encourage the use of the Siblings TEAKit among siblings, other interested groups including parents and/or caregivers and healthcare professionals will be asked to provide feedback on the Siblings TEAKit. Parents and/or caregivers and healthcare professionals will be recruited for the focus groups where they can can share their insights into the wording of questions for when siblings initiate conversations about their roles, and how the toolkit might be used in different settings such as at home and in health appointments.

Prior to the focus groups, participants will be asked to review the prototype of the Siblings TEAKit. This review should take approximately 30 min. During the focus group, participants will be asked to provide feedback on the relevance of the content in the Siblings TEAKit, as well as the barriers and facilitators to promote the use of the Siblings TEAKit among siblings.

Some examples of prompts and guiding questions are:

- Content: What information did you like? What information did you find helpful?
- Design: What did you like or not like about the design of the Siblings TEAKit?
- Use: How might the Siblings TEAKit be used? What might be the challenges with having siblings use the Siblings TEAKit?

The complete focus group script can be provided upon request to the first author (LN).

The first author (LN) and SibYAC member(s) will facilitate the workshop and conduct the focus groups. To promote engagement from participants during the workshop and focus groups, participants will have the option to share their perspectives verbally and/or type in the chat box. We will explore software, such as Canva whiteboards, in which participants can either type or draw ideas during the sessions.

The first author (LN) will write field notes during the workshop and focus groups. These field notes will include details about the date and time of the workshop and focus groups, how to establish rapport with participants, and perceptions about verbal and non-verbal communication from participants. Following the workshop and focus groups, the first author (LN) will write reflexive memos to reflect on the process of conducting the workshop and focus groups, as well as key findings that were mentioned by participants [61].

Implementation science frameworks

To design the questions and activities in the workshop and focus groups, we will use principles from implementation science frameworks. Implementation science involves the scientific study of methods to promote the uptake of research findings and evidence-based practice in health services [62]. The principles of implementation science will be used to inform the design of the toolkit to support siblings to achieve their target behaviour in initiating conversations about their roles, as well as to understand how the toolkit could be implemented in different settings. Namely, we will adopt the Capability, Opportunity, Motivation towards Behavior change (COM-B) model [63] to identify the key barriers and facilitators (Theoretical Domains Framework [64]) involved in adopting and using the TEAKit by families and healthcare providers, the core elements of the intervention, the elements that can be modified, and to describe the essential characteristics involved in the implementation of the TEAKit by families and healthcare providers (targeted behavior change). We will also apply the Consolidated Framework for Implementation Research (CFIR) Model [65, 66] to understand the factors involved in different settings for where and how the toolkit could be implemented. Figure 1 provides a visual diagram of the application of the approaches and theoretical frameworks used in this study. Further details about the use of these implementation science frameworks are described in the data analysis section.

Data collection and sample

Participants

Purposeful maximum variation sampling [67] will be used to obtain diverse perspectives of participants who represent different interest groups. Specifically, we will seek out participants of different ages and genders, in different sibling birth order (older/younger) in relation to their sibling with a disability, and invite those from different Canadian provinces, living in urban and rural communities, with different citizen status (e.g., immigrants, permanent residents, refugees, Canadian), Indigenous and First Nations, and French and English speaking as

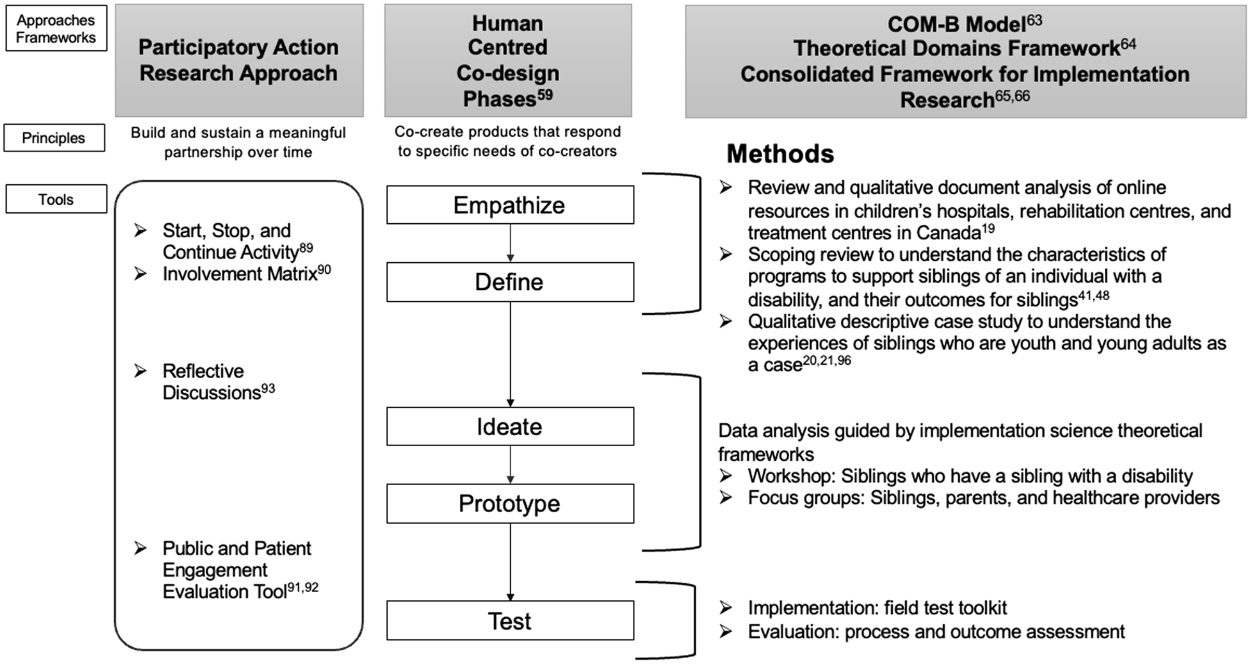

Fig. 1 A description of approaches and frameworks applied in this study

first, second, or third language. Currently, the workshop and focus groups will be conducted only in English as the resources available to this team do not include the capacity for translation into French during the conduct of this study. We plan to translate the final product of the Siblings TEAKit into French and potentially other languages.

We will recruit the following types of participants based on the inclusion criteria:

Siblings Participants will be recruited for the co-design workshop and focus group if they:

1. are a sibling of an individual with a disability (including physical and/or intellectual disabilities)
2. are between the ages of 14 to 25 years old
3. speaks English
4. live in Canada

Parents and/or caregivers Participants will be recruited for the focus group if they:

1. are a parent or caregiver of at least two children, one child with a disability (physical and/or intellectual disabilities) and one child with no known diagnosis of a disability
2. speaks English
3. live in Canada

Healthcare professionals Participants will be recruited for the focus group if they:

1) are healthcare professional with at least 5 years of experience working with children and youth with disabilities
2) speaks English
3) live in Canada

Participants will be recruited through childhood disability organizations and networks, including the CHILD-BRIGHT Network (headquartered at the Research Institute of the McGill University Health Centre), CanChild Centre for Childhood Disability Research at McMaster University, and the McMaster Autism Research team (MacART) at McMaster University. These organizations will also be asked to share the recruitment email and recruitment poster through social media, including the CHILD-BRIGHT website and Twitter, as well as, the CanChild website, Facebook page, and Twitter, and the McMaster Autism Research Team (MacART) website and Twitter, and Jooay Twitter. Our research team will also share the recruitment materials on social media (e.g., through Twitter and Facebook). Healthcare professionals will be recruited by email from the Alberta's Children's Hospital. Snowball sampling will also be used, in which participants could refer to other potential participants who may be interested in participating [68–70].

All participants will be asked the following screening questions by email. The sample size of the workshop and each focus group will have up to 6 participants for a total sample size of 24 participants who have experience to provide feedback for the Siblings TEAKit based on their knowledge related to family planning and discussions of sibling roles. This sample size has been shown to be sufficient to have a diverse range of perspectives with specialized knowledge and experiences that can be shared [71–73]. Further, small focus groups will allow for participants to interact with other with detailed and rich discussions to inform the design of the Siblings TEAKit prototypes [74]. There will be one workshop and three focus groups in total. The workshop will be a starting point to co-develop the initial prototype Siblings TEAKit, and the focus groups will allow for the iterative refinement of the toolkit.

This study has obtained ethics approval from the McGill University Institutional Ethics Review Board and the Centre for Interdisciplinary Research in Rehabilitation in Montreal, Quebec, Canada. All participants will be asked to provide consent prior to their participation.

Data analysis

Analysis of workshop and focus groups using implementation science principles

Implementation science principles will be applied to analyze the data collected from the workshop and focus groups. Descriptive statistics will be used to report and analyze the sociodemographic characteristics of participants. The audio recordings from the workshop and focus groups will be transcribed verbatim for directed content analysis [75]. The transcripts, field notes, and reflexive memos will be imported into the data management software, NVivo. The transcripts will be reviewed and analyzed in an iterative process. Inductive coding will be conducted to identify the key topics raised by participants in the interviews and focus groups. Deductive coding will be guided by the components of the COM-B model [63], domains of the Theoretical Domains Framework [64], and components of the Consolidated Framework for Implementation Research (CFIR) Model [65, 66]. The Siblings TEAKit will target the behaviour of the siblings to initiate conversations with their family to plan and prepare for their roles and the purpose (also known as the intervention function) of the Siblings TEAKit. Figure 2 outlines the steps for data analysis.

1. COM-B model [63]: This model will be used to understand the target behaviour in context and identify techniques to change the behaviour, and is comprised of three components:
   a. Capability: An individual's physical or psychological ability to perform the behaviour.
   b. Opportunity: A physical or social environment that is conducive for the behaviour to occur.
   c. Motivation: An individual has the intention to engage in the behaviour.

2. Theoretical domains framework [64]: The theoretical domains framework is a separate framework that complements the COM-B model by expanding on the details of the three components of the COM-B model through fourteen domains (see Table 1, adapted from [76, 77]). The domains of this framework can provide a more detailed explanation about the factors that can affect the target behaviour of the siblings.
3. Consolidated Framework for Implementation Research (CFIR) model [65, 66]: This model is comprised of five components: the innovation, inner setting, outer setting, individuals involved, and implementation process. Each of these five components includes constructs identified in Table 1 [66].

The COM-B model [63] and Theoretical Domains Framework [64] will both be used to understand the function of the Siblings TEAKit as an intervention to support siblings in achieve their target behaviour to initiate conversations about their roles. The COM-B framework [63] will be used to identify the broad components of the toolkit while the Theoretical Domains Framework [64] will inform the specific domains and details about the functions of the toolkit. The CFIR model [65, 66] will further complement these frameworks to analyze the focus groups and consider the characteristics of the Siblings TEAKit prototypes as an innovation on how to implement the Siblings TEAKit into different settings in the future. We will also consider whether there are individuals who may have different roles to implement the Siblings TEAKit as an innovation in different settings (e.g., facilitators, leaders, deliverers), and begin to identify the steps that may be taken in the implementation process in the future (e.g., tailoring strategies).

Following the coding of the transcripts from the workshop and focus groups, the behavioural change techniques that are commonly used to address each coded domain will be identified [78]. These behavioural change techniques can help to identify the key functions of the Siblings TEAKit, for example, to provide education to increase knowledge or to offer training to increase capability. The codes will also allow us to develop a comprehensive understanding about the factors in different settings, and the steps towards planning the process of where and how the Siblings TEAKit could be

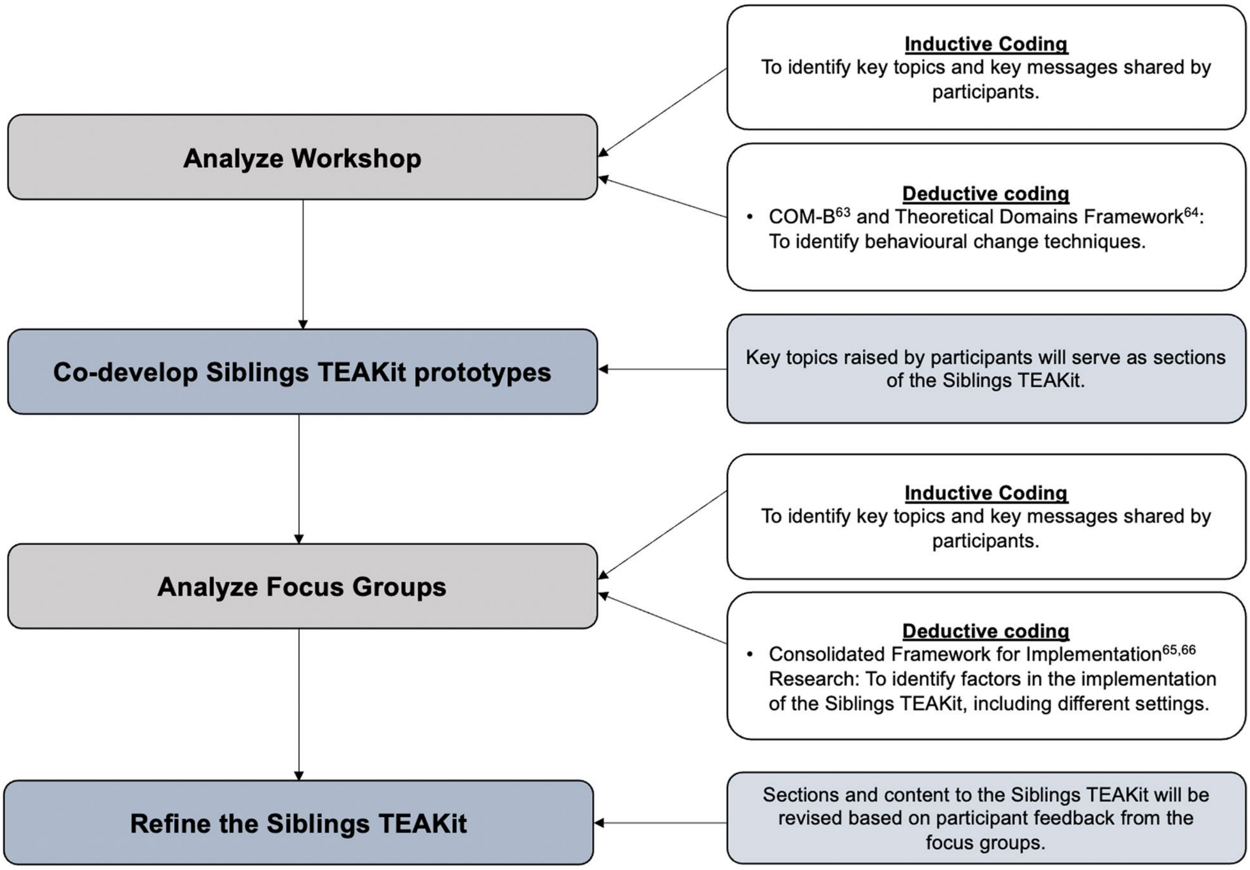

Fig. 2 A diagram to describe the steps in the data analysis process

implemented. The team will meet regularly to discuss and incorporate changes to the prototypes of the Siblings TEAKit based on the findings from the workshop and focus groups. Our multidisciplinary team includes individuals from different backgrounds including cognitive psychology, education, occupational therapy, physiatry, rehabilitation, user experience design, health policy, and patient-oriented research. We will develop the toolkit in an iterative process with our team, aligning with a human-centred co-design approach [59], where we will begin with drafting the content based on the identified codes and key topics raised by participants, and continue to refine the draft content with our team. The key topics raised will serve as the different sections of our toolkit. Our team includes members of the SibYAC who will be involved with drafting the content that draws from their lived and living experiences as a sibling. We will work with a graphic designer to compile our content into a toolkit format.

A lay summary of findings and Siblings TEAKit (version completed at the end of this study) will be shared with participants and the community as a form of knowledge dissemination. Findings will also be disseminated through conferences, publications, webinars, and our project study website [55].

Iterative evaluation of our partnership

It is important for the development and sustainability of research partnerships to align the principles of participatory action research and patient-oriented research. According to the Canadian Institutes of Health Research under the Strategy for Patient-Oriented Research, the guiding principles of patient-oriented research includes being inclusive to integrate different perspectives in research, provide support and flexibility to partners, having mutual respect for all partners, and co-building research together from the beginning [79]. Similar principles have been identified in patient-oriented research approaches in systematic and scoping reviews [50, 80–85], and specifically when conducting research with young people as partners [82–84, 86, 87]. To continue to strengthen our partnership in this study that reflects the principles of participatory action research and patient-oriented research approaches, we will use a developmental evaluation approach to reflect, evaluate and improve our partnership [88]. This approach will

Table 1 Description of the application of implementation science frameworks linked to each project objective and human-centered co-design phases

Project Objective	**Human-centered Co-design** [58]	**Application of Implementation Science Frameworks**	
		COM-B Component [63]	**Theoretical Domains Framework Constructs** [64]
To co-develop an evidence-based toolkit to provide education and training to support siblings in their roles	Ideate: Workshops	Capability	Knowledge
			Cognitive and interpersonal skills
			Memory attention and decision processes
			Behavioural regulation
		Opportunity	Social influences
			Environment context and resources
		Motivation	Social professional role and identity
			Beliefs about capabilities
			Optimism
			Intentions
			Goals
			Beliefs about consequences
			Reinforcement
			Emotions
		Consolidated Framework for Implementation Research [65, 66]	
		Components	**Constructs**
To further refine the prototypes of the toolkit	Prototype: Focus groups	Innovation	Source
			Evidence-base
			Relative advantage
			Adaptability
			Trialability
			Complexity
			Design
			Cost
		Inner setting	Structural characteristics
			Relational connections
			Communications
			Culture
		Outer setting	Critical incidents
			Local attitudes
			Local conditions
			Partnerships and connections
			Policies and laws
			Financing
			External pressure

Table 1 (continued)

Project Objective	Human-centered Co-design [58]	Application of Implementation Science Frameworks	
		Individuals involved	High-level leaders
			Mid-level leaders
			Opinion leaders
			Implementation facilitators
			Implementation leads
			Implementation team members
			Other implementation support
			Innovation deliverers
			Innovation recipients
		Implementation process	Teaming
			Assessing needs
			Assessing context
			Planning
			Tailoring strategies
			Engaging
			Doing
			Reflecting and evaluating
			Adapting

allow us to identify our partnership of where we were in the past, where we are in the present, and where we might want to go in the future. Further, this approach emphasizes ongoing reflections throughout the partnership and we will use the following tools to guide our reflections:

1. Start, stop, and continue activity: each SibYAC member will be asked to discuss their experiences about the activities being conducted using the prompts of: What activities should we start doing? What activities should we stop doing? And what activities should we continue doing? [89].
2. Involvement matrix: This tool will be used to ask each SibYAC member about the role(s) that they would like to have in the study; for example, a listener where they are given information, a co-thinker where they are asked to give an opinion, an advisor where they give (un)solicited advice, a partner where they work as an equal partner with the team, or a decision-maker where they take initiative and make the final decision [90].
3. Public and Patient Engagement Evaluation Tool (PPEET): Each SibYAC member will be asked to complete the PPEET to share their perceptions of their level of involvement in this study. The PPEET has undergone extensive usability testing and evaluation among patient partners and professionals from healthcare organizations [91, 92]. This study will use the participant questionnaire and project questionnaire to evaluate our engagement process.
4. Reflective discussions: As this study will use a co-design approach to develop the Siblings TEAKit, reflective discussions will be held to inform the co-design process. These reflective discussions will be guided by questions developed by a research team to discuss and evaluate how users are meaningfully engaged in the co-design process [93]. We previously described the importance of these ongoing reflective discussions to build and sustain our partnership over time [86].

Individual and group check-in meetings will be held with the SibYAC, guided by the discussion and completion of these tools. Monthly meetings will be held with the SibYAC to discuss updates and progress of this project. Figure 3 outlines the iterative process of how we will reflect on our partnership throughout the study phases of human-centred co-design.

Discussion

There are currently limited resources available to support siblings of individuals with a disability to have conversations about their roles with their family. This study draws on the lived experiences of siblings with lived experiences, specifically members of the SibYAC identified the need for this study based on their experiences of how they had to actively initiate conversations with their family about their roles. Members of the SibYAC reflected on how they would have liked to have had a resource to guide their conversations with their family. Similar findings have been shared in the literature, and siblings would like to have ongoing conversations about future care planning with their parents and family [34]. A SibYAC member also shared that she would have liked to be acknowledged by healthcare professionals for her role as a sibling [94].

This study design incorporates multiple strengths. First, the application of several theory-driven participatory action research approaches, implementation science, and human centred co-design to inform the co-development of the Siblings TEAKit as illustrated in Fig. 1 and as a general guide for other researchers in Fig. 4. This study was designed using a participatory approach from the beginning, where the relationship with our team of researchers and the SibYAC has been developed and sustained over time. Sustained relationships and trust with partners in participatory action research are important to contribute to the conduct, dissemination, and implementation of research findings [56]. The SibYAC was established in 2018 as an essential research partner and have been involved in several studies [16, 17, 19, 39, 48, 95] that informed the need to conduct this study to co-develop the Siblings TEAKit. In alignment with participatory action research principles [52], it was important that we develop a trusting environment where the SibYAC and researchers could have critical dialogues about ways to empower siblings in their roles. Our ongoing partnership with the SibYAC through multiple projects over the past six years allowed us to reflect on our expertise and experiences to inform the need to conduct this study, and inform each stage of the human-centered co-design to co-create the Sibling TEAKit.

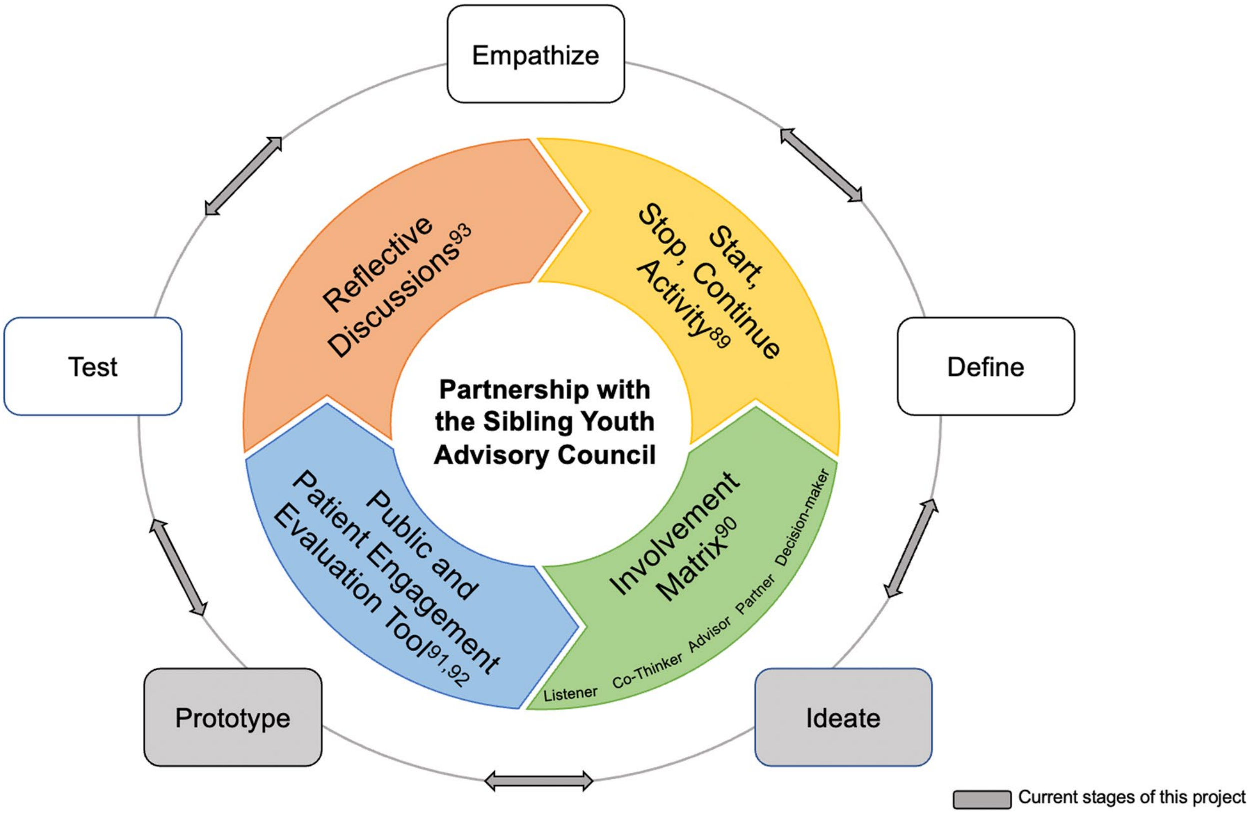

Fig. 3 A diagram to describe the iterative process of the evaluations of our partnership throughout the human-centered co-design study phases

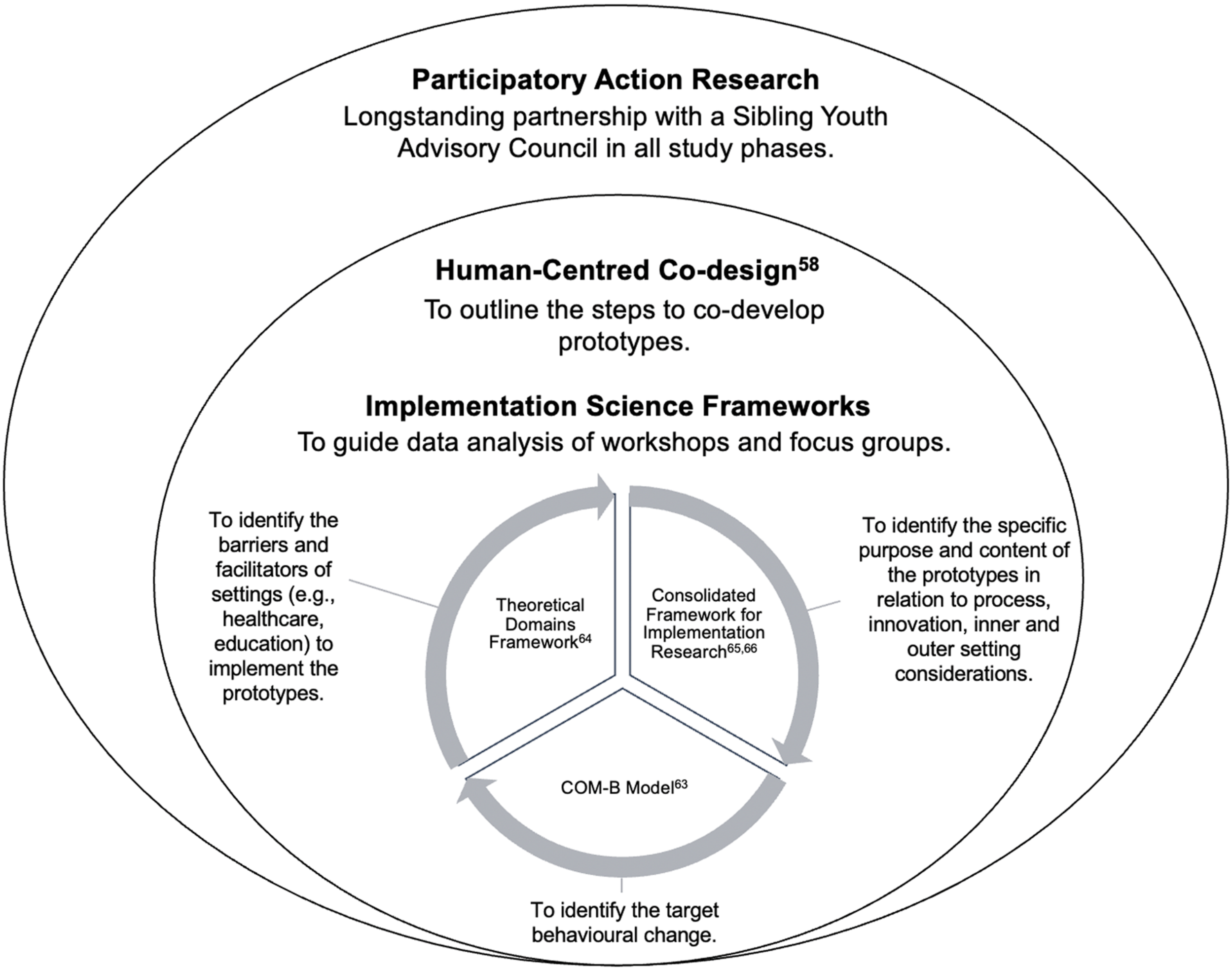

Fig. 4 A diagram to broadly describe the application of approaches and frameworks in participatory action research

A strength of this study is the intentional application of multiple theoretical frameworks of implementation science specifically, the COM-B model, Theoretical Domains Framework, and CFIR model to inform the steps of this research program to first co-develop the Siblings TEAKit and later implement this toolkit in different settings. A human centred co-design approach provided a guide to co-develop this toolkit with multiple interest groups, including youth and young adult siblings of individuals with a disability, parents/caregivers, and healthcare providers. The combination of approaches from multiple fields of study in a systematic manner will strengthen the co-development of a toolkit that could benefit the multiple interested parties targeted in this project: siblings of children with disabilities, families, and healthcare professionals.

A further strength of this study is the evaluation of our engagement with the SibYAC over a period of time. Evaluations of partnerships is needed to measure the impact of engagement with individuals with lived experience [96, 97]. These impacts can occur at different levels, including the value that the partnership brings to partners, researchers, and the studies and projects [86, 96, 97]. While there is no one size fits all approach to evaluating partnerships, reflections can be helpful to share learnings about how researchers and partners engage in research, as well as how their knowledge, attitudes, and thinking changes over time [97]. Over the past six years, our team has had ongoing reflective discussions using tools including the ‘Start, Stop, Continue’ activity [89], Involvement Matrix [90], and PPEET [91, 92]. These tools were beneficial to sustain and strengthen our partnership, for example, to understand the goals of each SibYAC member and how they would like to be involved in this study. We have had conversations with the SibYAC about the roles that they would like to have this study. SibYAC members preferred different roles, where some preferred to be a Co-thinker or Advisor where they provided feedback, while others preferred more active roles a Partner such as being a co-facilitator of the workshops and focus groups in this study. Our team is opened and flexible to new and changing roles that the SibYAC might have as we move forward with conducting this study. We will continue to incorporate feedback and strengthen our partnership based on our reflective discussions. Lack of evaluation and over time effects is a known limitation of participatory research approaches [54, 56, 98]. A solid evaluative framework will contribute to a better understanding of participatory approaches in implementation science.

This study will incorporate diverse perspectives from youth and young adult siblings with lived experiences, parents/caregivers, and healthcare professionals in the co-development of the Siblings TEAKit. Considerations about age, gender, birth order of siblings, type of disability, and ethnicity of the siblings will be taken into account. For example, female siblings who are older than the sibling with a disability often plan to take on caregiving responsibilities to their sibling with a disability [31, 32]. Cultural values may also influence the roles and responsibilities that siblings have. For example, certain cultures place high value on filial responsibility to the family and siblings may choose to prepare for their role as a future caregiver to their sibling with a disability [99–101].

While the intended users of the Siblings TEAKit will be youth and young adult siblings, the toolkit can provide support for meaningful conversations with siblings, parents/caregivers, and healthcare professionals. Parents may be reluctant to have conversations about the roles of siblings due to not wanting to burden the siblings about specific responsibilities, such as caregiving responsibilities [34, 35]. The ongoing discussion and negotiation of care responsibilities can be complex and emotional for the whole family, and this complex process should be recognized in the delivery of family-centred care [34]. The perspectives of parents/caregivers and healthcare professionals are essential and will generate useful insights about how this toolkit can be implemented in different settings, such as at home and in a healthcare setting, to have ongoing conversations about sibling roles. Healthcare professionals could ask questions to prompt families to have ongoing conversations about the roles of family member and share the toolkit as a resource. In other fields, such as pediatric cancer, healthcare professionals have identified a similar need to support young siblings by connecting them with hospital-based services and community resources [102, 103]. The Siblings TEAKit is a starting point to initiate conversations with families that is ongoing and so, the toolkit can be used by siblings throughout the lifespan as their roles can change over time [17, 20]. The Siblings TEAKit could also be personalized as a living document where siblings, families, and healthcare professionals could identify local health and community resources to refer to.

A limitation of this study is that the workshop and focus groups will be conducted only in English as the resources available to this team do not include the capacity for translation into other languages during the conduct of this study. We plan to translate the final product of the Siblings TEAKit into French and potentially other languages at the end of this study. We will also reach out to other health organizations and initiatives in Canada that are conducting transcultural validation of health research content to diverse languages and communities. We will also conduct knowledge dissemination by sharing the results of the study and the final product of the Siblings TEAKit through traditional formats including publications, conference presentations and webinars, as well as non-traditional formats by posting lay summaries on our study website [55] and on social media such as Facebook and Twitter.

Conclusion

Childhood-onset and developmental health conditions have an impact on the entire family system. Siblings are an integral part of family dynamics and may have unique roles in the process of caring for their siblings with a disability or chronic health condition. As such, they need to be informed, prepared, and empowered in their roles that can range from being a role model/mentor, advocate, supporter to a legal guardian and caregiver. This study protocol builds on knowledge and experiences from prior research on the roles of siblings in the care dynamics of young people with disabilities, and describe the theoretical and operational frameworks to create a toolkit that can support siblings to initiate ongoing conversations about their roles over time with their family and among different health and education professionals. Future studies can evaluate the implementation and effectiveness of this toolkit in different healthcare, rehabilitation, and education contexts, as a support tool for families. We also offer a framework for a structured iterative evaluation of research partnerships with diverse interested parties, advancing our knowledge of best practices for sustainable and meaningful research engagement with individuals with lived and living experiences.

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Authors' contributions

All authors contributed to the conception and design of this study. LN drafted the manuscript. All authors read, reviews, and approved the final manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

This study has obtained ethics approval from the McGill University Institutional Ethics Review Board (A03-E12-23B) and the Centre for Interdisciplinary Research in Rehabilitation (MP-50–2023-1839) in Montreal, Quebec, Canada.

Consent for publication

Not applicable.

Competing interests

The authors declare no competing interests.

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