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Interdisciplinary approaches to address health disparities for intellectual and developmental disabilities from underserved communities

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Interdisciplinary approaches to address health disparities for intellectual and developmental disabilities from underserved communities

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Editorial: Interdisciplinary approaches to address health disparities for intellectual and developmental disabilities from underserved communities

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Editorial on the Research Topic
Interdisciplinary approaches to address health disparities for intellectual and developmental disabilities from underserved communities

Individuals with intellectual and developmental disabilities (IDD) account for a significant proportion of the population with about 18% having any type of developmental disability and about 1% with intellectual disabilities globally (1, 2). Prevalence in low- and middle-income countries is even higher (2), and those with IDD are often considered the largest global minority group (3). Unfortunately, health outcomes are consistently worse for those with IDD compared to those without IDD. Many decades worth of data consistently show higher rates of mental illness, addiction, obesity, in-patient care use, and all-cause mortality (4, 5) for those with IDD. Additionally, those with IDDs tend to experience increased social isolation (6), family stress (7), stigma (2), and education barriers (8). These challenges are likely even more extensive for children with IDD from underserved groups (e.g., rural and inner-city children, racial and ethnic minorities) (9–11). Though the needs of individuals with IDDs are often unique and complex, care teams serving those with IDD largely work in isolation from one another (e.g., education vs. healthcare). As a result, the public health burden due to IDD is high with few coordinated services that are often structurally disconnected by policy or funding siloes (12). For example, individuals with IDD and their families must navigate complex and fragmented networks of services, which limits providers' ability to gather a full understanding of the individuals they are serving.

Despite this high prevalence and public health burden, individuals with IDD still often struggle to live independently. Moreover, the approaches used to improve the lives of those with IDD are often designed to work within isolated disciples or for contained populations, which limits opportunities for innovation. Therefore, there is a tremendous need to support interdisciplinary work that develops novel approaches to define, measure, and intervene in order to improve outcomes for those living with IDD across the lifespan, and their families.

This collection focuses on enhancing the understanding of complex systems of care by creating a forum for interdisciplinary teams that explore innovative solutions to a variety

of problems facing those with IDD. "Interdisciplinary" is broadly defined here and includes collaborative, family-centered approaches leveraging multiple systems. Collectively, these papers underscore a global commitment to advancing systems of care, professional training, and meaningful inclusion for individuals with IDD across the lifespan and address a wide range of critical areas of investigation. For instance, Ramachandran et al. review barriers to transitioning from pediatric to adult care for people with IDD, including limited provider training, poor care coordination, and lack of self-advocacy support. These authors recommend structured transition programs, enhanced provider education, and system-level reforms to ensure continuity and equity of care.

Three articles offer global perspectives. First, Zhong and Wijesinghe examine caregiving experiences among parents of children with cerebral palsy in Southwest China. They note that caregiver burden is influenced by child functioning, social support, and education, and the authors advocate for stronger psychosocial and policy supports to improve caregiver well-being. Second, Bakuwa et al. investigate the feasibility of a caregiver-led training model for parents of children with cerebral palsy in rural Malawi. They found this approach to be promising but highlight social and role challenges while recommending context-sensitive peer-led models to address workforce gaps. Third, Ferreira-Vasques et al. report on Brazilian norms for the Griffiths Scales of Child Development, Third Edition (GSCD-III), highlighting the importance of culturally relevant developmental measures and provides normative data for clinical and research use in Brazil.

Three other articles address capacity building projects that offer innovative approaches to improving accessibility and workforce development. Specifically, Fisher et al. discuss how the Leadership Education in Neurodevelopmental and Related Disabilities (LEND) program improves trainee knowledge and skill of how to effectively include people with disabilities in their work. Weber et al. describes a large workforce capacity building project called Project SCOPE. It is a scalable ECHO-based training program to enhance providers build capacity to work with children impacted by prenatal opioid exposure leading to neonatal abstinence syndrome. This article describes increased knowledge of intervention techniques, and confidence in implementation, as well as how the approach leads to exponential improvement in workforce capacity. Finally, Dudley et al. explore experiences of individuals with IDD participating in research to better understand how to support inclusive IDD research practices. Participants valued inclusion but noted accessibility and communication challenges. The authors suggest that developing accessible materials, supporting individuals with IDD to be in co-researcher roles, and ensuring financial compensation promote genuine inclusion and will improve research.

Finally, systems-level insights from public health and pandemic response are addressed in the last two articles. Specifically, Canpolat et al. examined national trends in special needs reports for children over a five-year period, finding disparities in access to services based on gender, geography, and socioeconomic status. The authors advocate for stronger data systems and targeted policy interventions to reduce inequities. Gotto et al., on the other hand, analyzed experiences with COVID-19 testing among children with IDD to identify broader lessons for care delivery. They found that communication difficulties, sensory sensitivities, and lack of provider preparedness were barriers to COVID testing, whereas caregiver advocacy, flexible procedures, and staff training in behavioral supports facilitated testing. The authors argue that these lessons can be applied to support more inclusive and adaptable healthcare practices beyond COVID-19.

Overall, these articles represent a diverse range of topics and fields, address complex and persistent challenges, and provide unique insights into solutions that have broad applicability. Together, they promote practical and exciting models for collaboration, training, and inclusion that center individuals with IDD and their families as partners in research, policy, and service design. However, ongoing research is needed to further develop these, and other innovations, that address challenges facing those with IDD, along with provision of robust person and familycentered support programs, and clinical interventions from an interdisciplinary lens. As this field develops, the use of inclusive, person/family-centered and interdisciplinary approaches will find the best, most effective solutions, and will create exciting new paths to allow those with IDD to thrive in their own communities on their own terms.

Author contributions

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Conflict of interest

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

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"This can certainly work…": stakeholder perspectives of the feasibility of a caregiver-led training program for caregivers of children with cerebral palsy in a rural setting in Malawi

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Introduction: Caregiver training is a key component of rehabilitation for children with complex lifelong disabilities such as cerebral palsy. However critical shortages of therapists in low- and middle-income countries like Malawi, reduce access to therapy. Introducing expert caregivers to assist with the provision of basic training on the condition for fellow caregivers offers a potential solution. However, there is a paucity of evidence regarding the implementation of such strategies in low-resource settings. The aim of this study was to explore perspectives of stakeholders regarding the feasibility of implementing a caregiver-led and delivered training program for caregivers of children with cerebral palsy in Malawi.

Methods: Over 5 days in January 2023, a caregiver-led training program, the "Malamulele Onward Carer-to-Carer Training Program," was conducted in Blantyre, Malawi. A South African master trainer traveled to Malawi and delivered the program to potential stakeholders including caregivers of children with cerebral palsy; physiotherapists; and community-based organization representatives. Stakeholder perspectives regarding the acceptability, demand, practicality and adaptation of the program were obtained through a combination of focus group discussions, in-depth interviews, and daily field notes. Data from the focus group discussions and in-depth interviews were analyzed using thematic analysis.

Results: The caregiver-led training program was deemed acceptable despite two areas identified as potential areas of concern; that the expert caregivers may cross practice boundaries and that their fellow caregivers may look down upon them. A demand for this program was expressed because of perceived relative advantages and relevance to caregiver needs. Participants indicated that the intervention could be easily delivered using local materials, absorbed and supported by existing community structures.

Conclusion: A caregiver-led training program offers an innovative way of supporting caregivers of children with complex disabilities such as cerebral palsy in low-resource settings. The stakeholder engagement demonstrated the positive perspectives of all stakeholders. The areas for modification and

adaptation highlighted by the stakeholders will be useful in strengthening the implementation of the program in Malawi.

KEYWORDS

implementation, caregiver-led, training, program, cerebral palsy, feasibility, Malawi

1 Introduction

Cerebral Palsy (CP) is a complex developmental movement disorder that presents with a variety of associated impairments, including epilepsy and cognitive difficulties (1). These disorders result in limited participation in activities of daily living (2). As a consequence of the lack of early therapeutic intervention and comprehensive lifelong rehabilitation, CP continues to cause severe disability in children in low- and middle-income countries (LMICs) (3). Currently, the global prevalence of CP ranges between 1.5 to 4 per 1,000 children (3, 4). The highest numbers of children with severe CP are in the rural areas in LMICs (5) leading to a significant health burden in these settings.

A population-based study conducted in two rural settings in Malawi reported CP as the commonest cause of motor disability, accounting for a quarter of all children with physical impairment (6). The study estimated that 2.1 children per thousand would benefit from physiotherapy services. However, access to early intervention and a continuum of care for children with CP is a challenge in Malawi with only 0.8 physiotherapists per 100,000 people; the majority of whom work in the four-government urban-based tertiary hospitals. A critical scarcity of rehabilitation services for children with CP, especially in rural areas therefore exists.

The complexity of CP requires frequent, regular and long-term therapeutic input to reduce disability (7). The caregiver needs to be knowledgeable about the specific type of CP their child has to ensure proper tailoring of therapies and adjustment of activities of daily living. For example, mobility, positioning and considerations for supportive devices vary depending on the type of CP the child has, its severity and the respective motor functioning levels (7, 8). In addition to this, the prognosis of the child's condition also varies and that knowledge is important for managing the caregiver's expectations, well-being and outlook on care needs (8). Moreover, children with CP vary in their participation abilities including play and can be challenged by the existence of other associated impairments including cerebral visual impairment (2). Provision of basic caregiver skills training in these aspects has been found to improve caregiver knowledge, skill and well-being and also functional outcomes of their children with CP (9, 10). It is the role of rehabilitation professionals to provide targeted caregiver training in all these aspects, however, workforce shortages in LMICs reduce their capacity to adequately train caregivers.

In line with the task-sharing strategy proposed by the World Health Organization (WHO), caregiver-mediated interventions have been recommended as an effective approach to harness the agency of caregivers to improve access to practical knowledge and skills in LMICs (11–13). These approaches focus on the engagement of primary caregivers of children with a disability, usually parents, and are commonly referred to as "parent-mediated/delivered" interventions (14). These programs are increasingly called "caregiver-led/delivered" approaches; which acknowledge that the child's primary caregiver may not always be a parent but could also be grandparents, aunts, uncles and siblings (15). In 2019, the WHO Caregiver Skills Training (CST) program was developed to support training and well-rounded support for caregivers of children with neurodevelopmental disabilities, specifically Autism, in LMICs (16). The WHO CST has been successfully adapted and implemented in several LMICs and is designed to be deliverable by non-specialists including "peer-caregivers" to enhance reach and scale up (17, 18). During the recent implementation of the WHO CST, these non-specialists have included community health workers, teachers and primary health workers. However, the engagement of caregivers themselves as program facilitators of the WHO CST is yet to be documented.

A South African non-profit organization, Malamulele Onward, championed the training of caregivers to work as peer facilitators (referred to as "expert caregivers" in this paper). In consultation with caregivers, the organization developed a training package, the Malamulele Onward Carer-2-Carer Training Program (MOC2CTP). Expert caregivers are trained to deliver a series of seven workshops to other caregivers of children with CP. The seven workshops impart practical knowledge and skills to caregivers which assists caregivers in understanding their child and the nature of CP. The workshops focus on teaching helpful caregiving practices which are integrated into daily life to enable each child to reach their potential while decreasing the burden of care. Over the past 7 years, the organization has trained over 50 expert caregivers in rural South Africa, Lesotho, and most recently Uganda (19). Qualitative studies involving over 400 caregivers who attended the program have demonstrated that the MOC2CTP results in increased caregiver understanding of both CP and their child; decreased feelings of isolation and self-blame; and feelings of hope and confidence in caregiving (20).

While caregiver-led and delivered training programs are being advocated and used for caregivers of children with various neurological conditions, including Autism and Zika-related congenital disorders (21, 22) there is limited work being done to formally study the feasibility of this approach specifically for caregivers of children with CP. Feasibility relates to the possibility of implementing an intervention or a program in a new setting. It involves assessing factors such as acceptability, appropriateness, relevance and practicality in terms of the availability of resources and time (23, 24). This means that considerations may vary depending on the specific end users of the intervention or setting of intervention. Therefore, establishing feasibility is necessary to ensure that a program is well-prepared for application within a particular practice setting to yield the intended results (24).

Abbreviations: CBO, Community Based Organization; MOC2CTP, Malamulele Onward Carer-2-Carer Training Program.

Recent feasibility studies that included caregiver-delivered aspects and also potentially children with CP have only focused on children at risk of neurodevelopmental conditions who were younger than 2 years (12, 13) and hence may not be relevant for older children with CP. There is a gap in the evidence regarding the feasibility of caregiver-led and delivered programs for children with CP. Moreover, in a setting like Malawi, where rehabilitation services are mainly led and delivered by physiotherapists, it is not known if this approach would be deemed acceptable, practical or adaptable to a local context. Therefore, this study sought to explore stakeholder perspectives regarding the feasibility of a caregiver-led training program, specifically the MOC2CTP for caregivers of children with CP in Malawi. Stakeholders refer to the end users; which are the caregivers and all the groups of people vested in the successful delivery of the program (25, 26); in this case, physiotherapists and the community based organization working with the caregivers.

2 Materials and methods

2.1 Study design

This was a qualitative study, using focus group discussions (FGD) and in-depth interviews (IDI) to elicit stakeholder perspectives. It comprises the first phase of a larger pilot implementation study.

The study design and tools were informed by Bowen's feasibility framework, which describes eight constructs for studying feasibility, *viz.*, acceptability, demand, practicality, adaptation, implementation, limited efficacy, integration and expansion (23). This study focused on four of these constructs: acceptability, demand, practicality and adaptation, as these were deemed relevant to guide the subsequent pilot implementation. The development of the data collection tools was informed by the Consolidated Framework for Implementation Research (CFIR) (27, 28) which provides tailored interview and focus group topic templates related to aspects of acceptability, demand, practicality and adaptation among other feasibility constructs (Table 1).

2.2 Study setting

The study was conducted in January 2023, at the Feed the Children rehabilitation center in Blantyre, Malawi. The rehabilitation center was well situated for hosting this initial study to accommodate the visit of the master trainer from Malamulele Onward who would introduce and conduct the caregiver-led training program.

Participants included members of the Tiyende Pamodzi Group; a community-based organization (CBO) in the rural community of Namwera in the Mangochi district, situated in the southern region of Malawi. Tiyende Pamodzi is a local non-profit organization which provides monthly one-on-one physiotherapy, access to assistive devices and nutrition support for children with disabilities. In January 2023, the center had over 400 children with physical impairments registered, with CP being the most common condition. The organization employs one physiotherapist to serve the 12 subsidiary centers within their catchment area.

Tiyende Pamodzi works with the Mangochi District Hospital, which like most of the rural district hospitals in Malawi, has a shortage of physiotherapists and rehabilitation assistants and consequently the reach of services for areas as remote as Namwera is limited. In 2023, the district hospital had one physiotherapist and four rehabilitation technicians servicing a population of about 1,323,159 (29). This setting therefore depicts a typical resource-constrained rural area in Malawi, yet with a well-organized community structure that can be a conduit for introducing successful innovation of a caregiver-led intervention (30).

2.3 Study participants

Participants comprised stakeholders of the proposed caregiver-led and delivered training program. They included 10 caregivers of children with CP, two physiotherapists, two executive committee members from the Tiyende Pamodzi Group CBO who were purposively selected. The investigator (TB) organized a meeting with

TABLE 1 Feasibility constructs informing the study interview and topic guides.

Construct	Description according to Bowen's Framework (23)	CFIR guide tools (27)	Example questions derived from CFIR tools
Acceptability	The reaction of participants and stakeholders to the training program & concept of a peer facilitator	Intervention characteristics: complexity (CIFR) Inner setting (implementation climate) (CIFR)	How complicated is the session/intervention? What is the general level of receptivity in your organization to implementing the intervention?
Demand	The perceived extent to which the new MOC2CTP is likely to be deemed necessary, utilized and received	Outer setting: patient needs (CIFR) Intervention characteristics: relative advantage (CIFR)	How well do you think the intervention will meet the needs of the individuals served by your organization? How does the intervention compare to other similar existing programs in your setting?
Practicality	The extent to which the MOC2CTP is delivered with the limited means, resources and circumstances of the context	Intervention characteristics: Design and packaging, cost (CIFR) Implementation readiness: self-efficacy (CIFR)	What is your perception of the quality of the supporting materials, packaging, and bundling of the intervention for implementation? How confident are you that you will be able to successfully implement the intervention?
Adaptation	Changes in the MOC2CTP that may have to be made for the intervention to accommodate contextual factors unique to Namwera	Intervention characteristics: Adaptability (CFIR) Inner setting: structural characteristics (CIFR)	What kinds of changes or alterations do you think you will need to make to this session/ intervention so it will work effectively in your setting?

the executive committee of the Tiyende Pamodzi to present the concept of the study. In consultation with Tiyende Pamodzi's physiotherapist, the executive committee chose the 10 caregivers; one from each of the 10 subcenters of the Tiyende Pamodzi group which would be involved during the implementation of the next phase of the study. Criteria for identifying caregivers were agreed upon by the research team and Tiyende Pamodzi executive committee. These criteria included being able to read and write, having at least 2 years of experience in primary caregiving of a child with CP and at least 1 year of involvement with the CBO, having a similar socioeconomic status as the majority of the caregivers, and having displayed qualities such as initiative, leadership, and empathy during community center programs. These attributes were necessary as it was important to start identifying caregivers who would later go on to be trained *expert caregivers*. Similar studies conducted in other LMICs have identified these traits as important for *expert caregivers* or *peer supporters* (20, 21).

The two executive committee members were selected based on their experience working with the CBO and their knowledge of its visions and goals. They also needed to have experience with the field work done by the CBO, empathy and a keen interest in learning. The two physiotherapists were selected based on their work experience with caregivers of children with CP, their interest in improving service delivery in rehabilitation care, and their willingness to participate in the subsequent implementation study. The study also gathered demographic details of the children with CP, including the type of CP and their level of functioning, using the Gross Motor Functioning Classification System (GMFCS). The GMFCS is a classification system that assesses the movement abilities and limitations of children with CP, including the need for assistive technology and wheeled mobility (1).

2.4 The training program

The Malamule Onward Carer-to-Carer Training Program (MOC2CTP) is a targeted skills training program which focuses on teaching caregivers to understand what CP is, identify the different types of CP, learn to problem-solving, positioning and handling skills related to each type of CP in the following areas; eating and drinking, play, communication and cerebral visual impairment. These aspects are embedded in blending therapy with activities of daily living and looking at CP management as a way of life. The main outcome of the program is improvement in caregiver knowledge and skills related to child positioning, feeding, mobilization, play and engagement, communication and vision. This is expected to improve child-level outcomes; including child-mobility, self-care, feeding, and social skills. The program also aims at improving caregiver well-being and quality of life.

The MOC2CTP was developed in collaboration with caregivers and designed to be delivered by caregivers of children with CP (*expert caregivers*) to fellow caregivers of children with CP in resource-constrained settings. It comprises seven workshops, each lasting between 2 and 2.5 h. Each session includes an ice-breaker activity based on themes of psychosocial support; information on a particular topic; a larger section of practical experiential demonstrations and as well as group discussions. Table 2 provides an outline of the content covered in the MOC2CTP.

TABLE 2 Content of the Malamulele Onward Carer-to-Carer program.

Module	Content
1	Introduction to CP: What is it and how does it affect my child
2	CP as a way of life: Looking after my child throughout the day
3	Getting active: Getting my child's body ready to move throughout the day
4	Eating and drinking: making meal times safe and comfortable for my child
5	Communication: My child and I understand each other
6	Cerebral Visual Impairment: Understanding where and what my child can see
7	Play: Unlocking my child's potential

2.5 Study procedures

A master trainer from Malamulele Onward in South Africa traveled to Malawi and delivered the seven-module training program to the study participants over 5 days. The master trainer is an experienced mother of a child with CP, who was initially trained to be a trainer by therapists at Malamulele Onward. She has over 7 years of experience in training peer facilitators in South Africa and also assisted in training expert caregivers in other countries including Uganda.

Drawing from Cornwall's´ participation model (31) the three groups of participants in this study participated at two levels to strengthen the assessment of the program; program recipient level and program observant level. The caregivers were the active recipients of the intervention, while the therapists and CBO representatives played the role of professional observers with some active participation as required. This allowed for co-learning and co-reflection about the training program.

2.6 Data collection

Data were collected concurrently with the training program through focus group discussions (FGDs) and In-depth Interviews (IDIs). FGDs were conducted with the caregivers daily after each module was presented while IDIs were conducted with the physiotherapists and CBO representatives on the final day. It was necessary to conduct FGDs daily to ensure that caregivers recalled and shared module-specific perspectives regarding feasibility and in addition to the overall appraisal of the whole program which was conducted on the final day.

The primary author (TB) facilitated all FGDs and conducted all IDIs. A trained research assistant, a human and environmental health specialist (BSc) with 2 years of experience in qualitative data collection, was responsible for note-taking during the FGD and IDI. The research assistant also took notes during the debriefing session conducted by the master trainer on the final day. The focus of the debriefing session was on providing feedback related to the delivery of the program to the Malamulele Onward program developers. This was different from the focus of the FDGs and IDIs whose focus was exploring aspects related to the feasibility of the program in the new context.

2.6.1 Focus group discussions

The daily FGDs were audio recorded and key points were captured and populated on a flip chart as the feedback continued to build each day. On the final day, the flipchart paper was displayed on the wall to prompt a reflective discussion with the caregivers regarding the program as a whole. The FGDs with the caregivers were conducted entirely in the local language, Chichewa. The daily post-session FGD with caregivers, took an average of 30 min while the FGD conducted on the final day lasted approximately 1 h.

2.6.2 In-depth interviews

Two IDIs were conducted with the physiotherapists and CBO representatives on the last day of the five-day training program. The in-depth interviews were conducted in the form of homogenous dyadic interviews. This is a process whereby the two therapists were interviewed together, and the two CBO representatives were interviewed together. This approach ensured a co-construction of feedback and ideas, tapping from both their shared and unique perspectives about the study setting (32). The interviews were conducted in Chichewa; however, the therapists were free to express their responses in English. Thus, the data had a mixture of English and Chichewa. The dyadic interviews lasted between 45 and 60 min each.

Due to the pre-determined sample size for the study, cross-triangulation of perspectives between the three groups of participants was used was used to further explore new phenomena until conceptual maturity was reached (26, 27). Cross-triangulation was done on the last day and issues raised during the week by caregivers were cross-checked with the professional observers and rediscussed by caregivers in the last overall FGD (33, 34). The topic guides for the FGDs and IDIs contained similar questions specifically related to the acceptability, demand, practicality and adaptation of the intervention (Table 1). Prompts were added to guide the administrator and enhance the depth of responses.

2.7 Reflexivity

All three authors are physiotherapists. TB has a little over 5 years of work experience with children with CP and it was her first time working with the MOC2CTP. She was primarily involved in data collection as the principal investigator for the study. She had no prior relationship with the caregivers and CBO representatives involved in the study but had interacted with both physiotherapists involved in the study during undergraduate training and clinical internship program, respectively. WS and GS have extensive experience in working with children with disabilities, including CP, and have conducted research in the field. GS led the development of the MOC2CTP and has over the years sought to understand how the program would work in other local and regional settings.

The authors reflected on their prior understanding throughout the research processes and were mindful to let the analysis be data-driven. Their experience enriched the depth of analysis and reflective enquiry.

2.8 Data management and analysis

The audio-recorded data were transcribed verbatim, in the original language which was mostly Chichewa (and some English) using Microsoft Word. The data were then translated into English and imported into NVivo software for management and analysis.

The analysis of the data was theoretically driven by Bowen's feasibility framework (23). Four constructs from this feasibility framework were used as global priori deductive thematic areas. Constructs from the CFIR were also used to describe organizing themes. An iterative process was followed during data analysis to derive preliminary emerging impressions from the data. This was followed by systematic coding of the data, using both inductive and deductive analysis. The data were initially analyzed inductively, where codes were generated as the researcher read through all sections of the data line-by-line assigning codes to segments of the data as they unfolded. These codes then underwent a second level coding whereby the original codes were reanalyzed deductively through categorization into basic themes that fed into the four pre-selected concepts of Bowen's feasibility framework (35, 36). Co-investigators checked for quality and agreement in codes.

2.9 Ethical considerations

Ethical approval was obtained from the Human Research Ethics Committee (Medical) at the University of Witwatersrand in South Africa (M220924) and the College of Medicine Research Ethics Committee at the Kamuzu University of Health Sciences in Malawi (P.04/22/3608). Written informed consent was obtained from all participants.

3 Results

Ten caregivers, two physiotherapists and two representatives from the community-based organization participated in the study. The community and district-level physiotherapists had approximately 2 years of experience working in the community and the district hospital, respectively. The CBO representatives had over 5 years of experience working with the CBO and working with children with disabilities and their families in the community. Table 3 describes the 10 caregivers who participated. Characteristics of their children are also described, including the child's level of movement function (denoted by "GMFCS"), and the type of CP the child presented with.

3.1 Qualitative findings

Several subthemes were identified under each of the priori deductive global themes (implementation construct), i.e., acceptability, demand, practicality and adaptability. Table 4 summarizes the basic themes arising from the data.

3.1.1 Acceptability

Acceptability was described in terms of intervention characteristics related to the intrinsic appeal of the intervention (perceived advantages and potential disadvantages of the program) and the general level of receptivity of the program by the CBO (implementation climate).

3.1.1.1 Intervention characteristics (advantages)

All participants expressed a positive impression regarding the nature of the intervention package and having fellow caregivers deliver

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TABLE 3 Description of the study sample.

ID	Caregiver			Child			
	Age	Relationship	Marital status	Age	Sex	GMFCS	CP subtype
Caregiver01	25	Mother	Single	6	Girl	4	Choreo-athetoid
Caregiver02	37	Mother	Divorced	3	Girl	5	Spastic
Caregiver03	43	Father	Married	5	Boy	4	Spastic
Caregiver04	28	Mother	Single	12	Boy	3	Spastic
Caregiver 05	24	Mother	Married	2	Boy	3	Spastic
Caregiver 06	38	Grandmother	Divorced	7	Female	5	Spastic
Caregiver 07	42	Mother	Widowed	15	Female	2	Mixed
Caregiver08	42	Aunt	Widowed	6	Female	4	Choreo-Athetoid
Caregiver09	37	Mother	Married	4	Male	5	Dystonic
Caregiver10	35	Mother	Divorced	2	Male	4	Dystonic

TABLE 4 Summary of themes identified for each thematic area.

Implementation construct	Organizing theme	Basic themes
Acceptability	Intervention characteristics (Advantages)	Knowledge of my child
		The program was well-organized
		Value in being taught by fellow caregivers
		Lots of learning
	Intervention characteristics (Potential disadvantages)	Caregivers looking down on the peer facilitator
		Caregiver facilitators not knowing their limits
	Implementation climate (positive)	Aligned with CBO goals
		Support structures available
Demand	Relative advantage	Complement to existing interventions
		Less burdensome to caregivers
	Patient needs met by program	Learning how to deal with practical issues faced
		Acquiring new knowledge
	Patient needs not met by program	Need to address toilet training issues
		Need to include pain and sleep disorder issues
		Need to include a component on child evaluation
Practicality	Intervention design	Complicated for the facilitator, needs time to prepare
		Some content difficult/complex needs more time to deliver
		Easy for recipients to grasp because of lots of pictures
	Cost	Easy to deliver using local resources
		Easy to absorb into existing community center activities
	Implementation readiness: self-efficacy of prospective facilitators	Easy to deliver to fellow caregivers
		Knowledge of things to share with friends
Adaptation	Adaptation (areas of the intervention needing change/improvement)	Need to find appropriate Chichewa names for CP subtypes
		Need to repackage the content on food types in workshop 4
		Need for introduction of other disabilities in children in Workshop 1

it. The basic themes regarding the perceived advantages of the program were: (i) knowledge of my child; (ii) the program was well organized, (iii) lots of learning and (iv) the value of being taught by fellow caregivers.

Caregivers felt that the program enabled them to know their children better and understand their needs and helpful ways in which they can provide care.

> *"Because what I have liked are the new things that we have learnt, of course, I already do most of the therapies. But I really did not know what type of CP my child has, but now I know that there are several subtypes and I know to which group my child belongs and I now know that he is a level 4. And most importantly I know that now I need to do a lot of these and these for him to reach another level of ability. I am so happy!"* Caregiver 04

The professional observers appreciated that the program was well organized and packaged. They felt that the material was delivered in a way that was easy to follow

> *"It was good in several ways for example the content was easy for the people to understand it was really, well prepared so that depending on the nature of the participants it made it easy for them to follow and it even made sense to me as a therapist that oh this is what is supposed to be done."* Therapist 01

> *"The master trainer was well organized and could do it so well we could tell she is experienced in doing this… and the pictures are good, as we can see the training used a lot of pictures which the caregivers could follow and relate to."* CBO representative 2

Caregivers also felt that the program provided room for learning new important lessons and practices.

> *"For me for all the lessons from Monday to Friday, I have found them to be really good because we did not know anything, we were just living with our children but we did not know what to do with them, how to enjoy the child, how to take care of the child and the type of CP that the child has but now we know the types, the levels of CP (narrates the different types and levels of CP)"* Caregiver 07

The concept of the program being delivered by fellow caregivers was also received positively in several ways. Caregivers felt that they would value being taught by a fellow caregiver who knows what they go through, fellow caregivers were perceived as easily approachable since they share the same language and are not strangers. Expert caregivers were also regarded as a potentially valuable support for therapists and would increase access to therapy for children.

> *"Whereas with me, I interact with these people even in the community, we greet and chat. They can ask me anything anywhere…Moreover, for me who has the same problem as them to approach them and give advice on things they can do with their child, they will definitely appreciate this advice and not look down on me."* Caregiver 04

3.1.1.2 Intervention characteristics (potential disadvantages)

Physiotherapists identified two potential disadvantages of the program: (i) that the expert caregivers may not know their limits, and (ii) that they may be looked down upon by their fellow caregivers.

Physiotherapists felt that expert caregivers may start practicing beyond the boundaries of their role in the program. The physiotherapists agreed that this would require a precise explanation of roles and continued supervision by therapists to prevent harm.

> *"It is really a problem that can arise. So, to me this problem it's a question of making sure, like you put it earlier that there is supervision from a therapist. And even there should be a clear distinction that this is the therapist and this is the trained facilitator, who has been taught to assist the therapists…So, there should be a balance. Frequent checks and balances."* Physiotherapist 02

The physiotherapists also felt that there was the possibility of caregivers looking down on the expert caregivers and slighting their role and work. On this aspect, both the caregivers and therapists acknowledged this possibility and identified "exchange center visits" as a potential solution alongside good attitude and cordial presentation of the expert caregivers toward their fellows, including personal resilience.

> *"One disadvantage that I see … what am trying to say is after they get this training won't they pose in the village and say am the therapist? How do we make sure that they should know their position? They should know their position that okay this is our limit if we go further than this, we may harm. So that the only other problem I see should be addressed."* Physiotherapist 01

> "We are organized in centers and we have lots of centers. We can arrange that maybe I should go and facilitate at her center (pointing to a fellow caregiver) and she may come and facilitate training at my center as a visiting facilitator. And because they will see that a visitor has come, they will give more attention and be interested to hear what the visitor has brought. Likewise, I can then go to her center and she will support me. I think that could also help in case we feel that we may be looked down upon us" Caregiver 01

3.1.1.3 Implementation climate

Two basic themes arose under implementation climate (i) intervention aligned with CBO vision and goals and (ii) support structures available.

CBO representatives felt that the intervention was well aligned with its goals and vision and hence were optimistic about its success and showed keenness to implement it.

> *"This intervention is well aligned to our goals and vision. We have already been making efforts to implement some of these things. Now that we have learnt a lot more, we will implement them better"* CBO representative 01

The committee representatives and physiotherapist from the CBO envisioned how the program would be easily supported by the already existing structure of the community of volunteers. They felt that community volunteers working with the CBO could also support the work of expert caregivers and make things work.

> *"In addition, we also have community volunteers across all centers who can support these expert caregivers. We have like 49 of them in total. Each area at least has 3 or 4 volunteers. We encourage these volunteers to visit the homes of children with disabilities monthly and write reports. So, in our organization, this can certainly work."* Physiotherapist 02

3.1.2 Demand

Demand was defined in terms of intervention characteristics related to the extent to which the intervention program was deemed useful and relevant in comparison to existing ones (relative advantages) and the extent to which the program meets the needs of the caregivers of children with CP.

3.1.2.1 The relative advantage of the intervention

Two themes arose related to the relative or comparative advantage of the intervention: (i) a complement to existing interventions and (ii) less burdensome for caregivers.

Participants felt the program would complement the existing physiotherapy services and that it would result in increasing access to frequent therapies therapy for their children.

> *"Lessons like these going to our area will be the first time. Although other interventions are going on, we will be adding more and us being the first, we may turn out to be an example for other areas and help spread the program to other areas. And we will be able to do more with our children at home!"* Caregiver 08

> *"We tell the caregivers morning afternoon and evening do some therapy but now with the season that we are in for example, they have to go to the farm, they just do not have time. They cannot get time to do therapies. But now they are being encouraged to incorporate therapy into ma activities of daily living of which they would not spend a whole day without doing these. So, I feel this one is feasible considering the setting that the caregivers live."* Physiotherapist 02

Caregivers felt that the program presented a way of life that would ease their caregiving roles related to implementing therapies at home. The blended approaches presented to them meant that they would not struggle to create extra time for implementing therapies, which they already struggled with.

> *"That is what touched my heart A lot because we weren't finding time sometimes but with what you have explained we have known that sometimes as we are doing daily activities like feeding or bathing the child, we can do helpful activities at the same time."* Caregiver 03

3.1.2.2 Needs met by the program

Caregivers indicated that the program addressed real needs that they had and their perceptions revolved around two basic themes: (i) learning how to deal with practical issues faced and (ii) acquiring new knowledge.

Caregivers appreciated that the program addressed practical caregiving issues such as feeding, positioning and communication, which are things that they had difficulties with.

> *"We really appreciate it because such a program as this is needed and needed urgently so that we may assist our fellow caregivers as soon as possible. I can say that all the problems that were identified through the lessons actually exist in our communities, we see them… the feeding problems, positioning our children poorly and even failing to communicate with them! And so, I feel that the program will practically help our fellow caregivers and also improve the lives of many children."* Caregiver 05.

Caregivers also identified several things that were new to them which they recognized to be very important and useful. Physiotherapists also acknowledged having learnt things that they had never learnt even during their professional training including the management of Cerebral Visual Impairment (CVI)

> *"Ah, the needs are there indeed. Coming here we have learnt a lot that we did not know. We have learnt about the different types of CP and levels of function of children with CP and much more which our friends back home do not know."* Caregiver 09

> *"Mmmh, in terms of communication I saw that it went well and today as well (referring to the CVI lesson) I also saw that it went well and to say the truth as a therapist I have also learned things that I have never covered before and if we apply them well it can help a lot. So basically, I think this program is useful. That is my overall impression."* Physiotherapist 02

3.1.2.3 Needs not met by the program

There were other important child-related needs that the professional observers felt were not addressed by the program: (i) toilet training issues (ii) pain and sleep disorders and (iii) child progress evaluation. They recommended the addition of these aspects in this or future programs.

The physiotherapists mentioned how toilet training was one of the issues that caregivers struggled with when taking care of their children.

> *"The issue of toilet training is one of the problem areas we are facing in the community centers we offer services. Most of them are very poor, they cannot afford to buy Pampers…You find a 12- or 14-year-old just lying down, a level perhaps 4, not communicating or showing signs whatsoever. They leave such a child at home and find the child in the evening. No one has ever told them about toilet training the whole of the child's 12 years of life. So, adding a toilet training component would be helpful."* Physiotherapist 02

The physiotherapists also remarked that pain, sleep disorders and spasticity management could be other areas that caregivers would benefit a lot.

> *"But then we could also look at issues of pain and spasticity and sleeping disorders. I don't know how they can be incorporated*

> *whether it should be the same expert caregivers or whether that would be giving them too much to handle.*" Physiotherapist 01

The CBO representatives remarked that assessment of the progress of children's activity and participation was important and would have been good to include in the program.

> *"Perhaps the one side that I did not see was the area of evaluation. How can we see that our child with CP is improving or changing? That is what I also discussed with the therapist that these areas were not touched.*" CBO representative 01

3.1.3 Practicality

Practicality was defined in terms of intervention design, cost and implementation readiness. The section that follows outlines the themes under each of these predefined constructs of practicality and the direct quotes representing participant perspectives, respectively.

3.1.3.1 Intervention design

There were 3 basic themes related to intervention design: (i) complicated for facilitator to prepare, needs adequate time (ii) some content complex, needs more time to deliver (iii) easy for recipients to grasp because of lots of pictures.

The professional observers felt that the program was quite complicated to put together and would require time for the facilitator to master

> *"Yeah, this time am speaking from the part of the person delivering the training, that it's complicated, and you have to take it from that point that you ought to prepare very seriously otherwise it cannot come out as good as the expert trainer delivered it. If not, well prepared you could end up confusing the participants. So, I was speaking on the part of the one delivering."* Physiotherapist 01

> *"Well, with pictures sometimes… someone else, like a caregiver selected to facilitate, they may interpret pictures differently and participants may fail to understand the things. The expert trainer was well organized and could do it so well because she is used to doing this. Perhaps our facilitating caregivers will need notes to guide them and more practice to be able to use the pictures like she did …"* CBO representative 02

Participants observed that some sections of the program were complex and contained some very new concepts that would require more time to understand.

> *"Today's lesson (module 3) was long and hard to understand in a short period. It has a lot of parts; wanting to differentiate that if this child is in this type CP this is how can you take care of him and how can we prepare him in other activities… it has a lot of sections that one needs to understand."* Caregiver 08

> *"If the content is spread over do one topic per day. For example, doing communication alone and not combining it with play. Then we would not say that the content is too much. During this week we have covered everything in five just to showcase the program. However, I feel that if it were delivered in 7 days as designed it would be good."* Physiotherapist 02

Participants acknowledged that the training program was easy for learners to follow, especially owing to the many pictures and practical demonstrations that were used to illustrate concepts.

> *"All sides were seen to be easy because of the pictures because we were quick to tell that this picture means that so we were quick to differentiate with the words but the pictures we were quick to capture the idea that when we get home and see a child like this at this stage, I will explain to them like this. That is what I have seen."* Caregiver 08

> *"Maybe just to add on that, the content yes was a lot but because of the pictures and the practical demonstrations at least the participants could grasp a lot of things through the pictures and demonstrations. It's the one part of the program that made the program really good and made it simple for participants to grasp ideas easily."* Physiotherapist 02

3.1.3.2 Cost

There were two themes related to cost: (i) easy to deliver using local resources (ii) expert caregivers may expect something in return.

Apart from the printed pictures, caregivers felt they could be able to source all other materials to deliver the program

> *"If we take things from the village people will be quick to accept it. All of these materials we can find and make ourselves…at least except the pictures. If we take our local things, we will give them a picture that they can afford the things that they can manage to give the child and they are the things the child will be eating daily that they can find."* Caregiver 01

There was the worry that expert caregivers may not be willing to work without expecting anything in return. They therefore felt that this may affect sustainability. However, representatives felt that the time-to-time incentivization system that the organization followed could be effective for expert caregivers too. Moreover, the caregivers felt that they would not expect to be paid for sharing information and skills during their usual community center activities.

> *"The biggest issue is sustainability, we would want maybe to scale up so that we train more people so that we extend the program, so it is 10 then maybe next we can think of another sample so that the program can grow and reach every caregiver…not many people are willing to do this work without expecting anything in return are few."* Physiotherapist 01

> *"Our system is that although the volunteers are not paid, we still give the people incentives from time to time according to the needs that they get. For example, in times of hunger or famine they are supported…and even during the farming season. So, this is a system whereby they don't get paid, they know that they have somewhere to*

> *go when an emergency arises. But it's not like it's a pay but at least they know that being attached to the organization I will be assisted when I have a problem.* CBO representative 02

> *"Let me add on the same point, we will be sharing with our friends during our usual community center activities, we already share things when others get training like that nutrition training last time. We have learnt this time, then we will pass on to our friends, and those friends will pass on to others. We will not expect to be paid for sharing with friends in our centers."* Caregiver 09.

3.1.3.3 Implementation readiness: self-efficacy of prospective facilitators

Caregivers felt that following their participation in the workshops during the week, they could be trained as expert caregivers and deliver the program as facilitators. They expressed this in two ways: (i) the material would make it easy to share with fellow friends and (ii) they understood the knowledge and skills to share with friends.

The practical nature of the program made caregivers feel that they would easily be able to deliver the program to their fellow caregivers.

> *"On my side, I would not have problems delivering the training because of the skills that I have received. Also, the materials that were used were really helpful and it's those pictures which aided understanding and also remind you as a facilitator to remember things that you may have missed."* Caregiver 06

The caregivers felt confident of their understanding of the material and, their ability to share the knowledge and skills with their fellow caregivers

> *"It will not be difficult to teach them because most of the things we will be delivering are things that we have already covered this week. Moreover, the people we are going to teach are not strangers but women whom we interact with daily so it will not be difficult."* Caregiver 02

3.1.4 Adaptation

Three possible areas for adaptations arose: (i) need to find appropriate Chichewa names for CP sub-types, (ii) need to repackage the content on food types in the workshop 4 and (iii) need to include an introduction to other disabilities in children in workshop 1.

Participants recommended that the CP subtype names have better and consistent local translations that will be practical for use during the program implementation.

> *"Yes, we are saying that most parts we understood easily because they were all in Chichewa, the hard parts we found were those names that were in English and they may be difficult for our friends back home because of our literacy levels. Yes, so we would have loved if we had Chichewa translations for those sections too."* Caregiver 04

> *"Let's find good Chichewa names for the CP subtypes so that it is not difficult for people to identify and follow. Even for the pattern names, quad, hemi, Di. Let's also find proper names so that when asked the people should be able to name without problems."* Physiotherapist 01

Caregivers recommended repackaging the content on food groups in workshop four ensure consistency with what the caregivers learn at the local health facilities on nutrition. That is: to teach the six food type groups as is taught in Malawi instead of the three food type groups in the MOC-2-CTP manual.

> *"I was worried about the food groups. The way we group them and the way our friends are grouping them is different because we expand those groups into 6 groups and give them names while here, we have been given 3 names. That is where my worry was like how will I explain to my friends for them to understand".* Caregiver 08

Caregivers recommended the inclusion of a section that introduces childhood disabilities before narrowing it down to CP. This is because, in their community center programs, they deal with different types of childhood disabilities not only CP. Therefore, they perceived that adding a generic introduction would be beneficial.

> *"But I have one concern about a challenge that we might meet when we go back to our community centers. Because in our centers we have different types of disabilities amongst children … And so, I was thinking that in the beginning, you should have explained to us about the different disabilities that are there and then tell us that this time we are focusing on CP."* Caregiver 01

4 Discussion

This study sought to explore the feasibility in terms of perspectives of stakeholders on the acceptability, demand, practicality and adaptation of a caregiver-led training program for caregivers of children with CP in a rural Malawian setting. The MOC2CTP was introduced in the Malawian setting for the first time for potential users to appraise before proceeding with implementation in the community. Our findings demonstrate that the intervention was deemed acceptable and that there is demand for such an intervention in this setting. Participants also felt that it would be practical to deliver the program in their setting but that it required contextual adaptations.

The MOC2CTP program was deemed acceptable by participants due to its practical learning opportunities, well-organized structure, and alignment with the CBO's vision. Participants felt that the program's activities were not demanding and would allow them to balance their daily activities in the rural setting. Similar positive perspectives were reported in other skills training interventions for caregivers in LMICs, such as the Juntos in Brazil, and Baby Ubuntu in Uganda (12, 13, 19) where the focus was learning and training through daily activities. In rural settings, interventions that integrate into caregivers' daily lives and provide peer support are more likely to be accepted. Caregivers prefer interventions that support their natural role and help them cope with daily activities rather than focusing solely on technical skills transfer.

Moreover, the concept of teaching and learning from peers was perceived as supportive, encouraging, and motivating. Caregivers felt that they would be easily accepted by fellow caregivers because of shared experiences, culture and language. These motivators have been identified in similar programs like in the Juntos in Brazil, where the experiential knowledge of the "expert mother" was highly esteemed by fellow mothers who felt encouraged and motivated (21). The engagement of a caregiver as a program facilitator enhances the provision of a family-centered rehabilitation service. Experiential peer support goes a long way to positively impact caregiver well-being beyond the improvement of practical skills related to caregiving.

However, a concern was raised regarding the risk of the expert caregiver crossing boundaries. Recent reviews on peer-led interventions for caregivers of children with neurodevelopmental conditions have not reported this concern (22, 37). However, in a study done in 2022 to investigate benefits and challenges of peer support programs, organizers were also concerned about intervention fidelity by "peer supporters" (38). They ensured careful selection of the "peer supporters," adequate description of their roles and limits and continuous support. Clear definition of boundaries during the training of facilitators and continued supervision have been highlighted as key to the successful integration of peer facilitation (14, 22, 38). There is evidence of successful and safe engagement of peer facilitators in skills training programs with high perceptions of acceptability reported. Mothers of children with Zika-related neuro-disability and children at risk of CP safely and successfully co-facilitated programs in the Juntos and Baby Ubuntu programs in Brazil and Uganda, respectively, (9, 17). Therefore, during the subsequent implementation of the MOC2CTP, boundaries need to be clearly explained and emphasized in addition to the provision of supportive monitoring and supervision.

All participants recognized this intervention as a great need and relevant to fellow caregivers in their setting. Limited access to a single physiotherapist in their area was a major concern, and this intervention was deemed helpful in addressing it. There is a persisting critical shortage of rehabilitation professionals in LMICs, including Malawi (39). Services in most LMICs are limited to urban communities and render rural populations deprived of services (20, 39). Caregiver skills training programs have been found to increase access to services for caregivers of children with CP and other neurodevelopmental conditions. This is mainly because caregiver skills training programs focus on the engagement of non-specialists which enhances access, affordability and availability of services in the community (17, 26). For example, in rural settings of Ghana, South Africa and Uganda, skills training programs like the Getting to Know CP (GTKCP), MOC2CTP and Ubuntu programs respectively, were the most accessible services to caregivers and any other services were hardly accessible (10, 13, 20). It is not surprising that the prospective stakeholders in this study have also recognized that the MOC2CTP would improve services and make rehabilitation more easily accessible.

Moreover, the CBO representatives and caregivers affirmed the practicality of delivering the program in their community. The CBO representatives felt the program could easily be integrated into their existing programs and that incentive system could also support expert caregivers and ensure the continuity of the program. Caregivers were also confident of their ability to deliver the knowledge and skills imparted to them to their fellow caregivers using resources in the community. Stakeholder readiness and the ability of programs to integrate into existing community programs has been identified as a key indicator of feasibility (13). Moreover, in this setting, the existence of community centers and ongoing caregiver support groups presents a platform for ongoing peer support and continued practice. Such a structure is important for sustainability of, unlike settings where organized support groups are not available (18).

On the other hand, the professional observers felt that although the materials and methods were easy for recipients to grasp., they would be quite complex for the expert caregiver to organize and deliver as intended. They recognized that it would take adequate time and exposure to the delivery kit for them to be able to use the teaching materials with their peers. This highlights the importance of adequate training for expert caregivers, which has also been emphasized by various authors on peer-led training (11, 20, 40). The recommendation for this particular training package is 80 h to achieve consolidation of skills. In addition to this, there were modules which caregivers felt that would require more time for their fellow caregivers to adequately understand, which was attributed to low literacy levels. Similar recommendations have been made in other caregiver training programs, emphasizing adequate oral discussion and the use of simple visual representations (18). Therefore, giving more time for facilitators to explain concepts using the pictures will be beneficial in the pilot implementation of this program.

The stakeholders recommended adaptations to the program related to language, information packaging and diversity of childhood disabilities at their community centers these modifications would ensure that the information is easier to grasp. For example, they recommended further linguistic translation of the CP subtypes in the local language, Chichewa and the inclusion of an introduction section that differentiates CP from other neuro-disabilities of children who come for service in the community centers. Participants also recommended including a description of the three food groups in the "eating and drinking" session to reflect caregivers' knowledge of the six food groups taught in health facilities in Malawi (41). This demonstrates the necessity of contextual adaptation of interventions before moving to the implementation phase of the larger study. According to the WHO, skills training programs for caregivers of children with developmental disorders like CP, should have materials that are linguistically appropriate and relatable (40). Therefore, these suggestions will be useful in subsequent adaptation of the training program before it is introduced to the community.

In addition to the areas for possible adaptation, stakeholders identified other needs which the current program did not address. These were toilet training and pain management, which are aspects not addressed by the current MOC2CTP. In a recent Ethiopian study, caregivers also identified toilet training as an area not addressed by the WHO CST (18). The need to address pain in children with CP has also been affirmed by the children, caregivers and clinicians in several studies in recent years (42, 43). In LMICs, addressing these issues is complex due to limited access to specialized interventions and uncoordinated delivery of available interventions which result in missed opportunities (39). While toilet training goals may not be feasible for all children owing to levels of severity it is worth investigating in future studies how the community platform may be utilized to start addressing these more specialized needs in a meaningful way.

Overall, the perspectives of the stakeholders regarding the feasibility of the training program were positive. They identified

several areas that needed to be modified and their suggestions were useful for ensuring contextual adaptation of the program. In preparation for the subsequent pilot implementation, the investigators engaged a team including four clinical physiotherapists, two social science linguists, the caregivers, the CBO executive and the Malamulele Onward organization to work on the areas recommended for adaptation. Conceptual translation of the CP subtypes was done through consultation with clinical physiotherapists, linguists and the caregivers involved in this study. Development of an introductory section on childhood disabilities was done in consultation with the program developers, the Malamulele Onward. This also included repackaging the section on food types in the "Eating and Drinking" module. This was done in consultation with the personnel from the community health center and the caregivers themselves. Considerations for slow-pacing the delivery of the content were made. A day was allocated only one module, to give room for extensive discussion of concepts and practice. This highlights the essential role of intervention stakeholders in providing critical perspectives important for the contextual adaption of innovations from other contexts.

5 Strength and limitations

This stakeholder engagement study drew from participatory action research ethos which propagates for active engagement of all stakeholders of proposed interventions at all interactive stages of implementation (44, 45). Inviting these groups of stakeholders or potential users, strengthened the study since it provided the opportunity to generate contextual knowledge in a co-creative way (44, 46). Secondly, a multi-methodological design qualitative design was applied (33, 34) with two different data collection methods, namely dyadic interviews and focus group discussions. This method took advantage of the strengths of both, as reported by Creswell (47) and augmented the trustworthiness of this study (32). For example, interviewing professional observers in a dyad enabled the co-generation of ideas between the professionals that would not be possible through separate in an individual In-depth Interviews.

Furthermore, there was triangulation of perspectives from all three sets of data to obtain a better understanding of concepts. For example, physiotherapists felt that a peer facilitator would be looked down upon by fellows, however when the researcher posed this concern to caregivers themselves their perspective was that being looked down upon would be because of the way they would present themselves to their peers. They highlighted the need to remain humble and empathetic to earn respect from their peers. This strengthened the rigor of analysis and interpretation and also complemented the researcher's reflexivity.

This study had an important limitation. The expert trainer only had 5 days to demonstrate the delivery of the intervention, which comprised seven workshops. It would have been ideal if each workshop and subsequent discussion were held on separate consecutive days rather than delivering seven workshops over 5 days, which meant that some of the workshops were combined into one session. Although the aim of this initial exposure to the program was for stakeholders to merely appreciate how the program is designed and delivered, the short period may have affected the perception of stakeholders. Lastly, the views of the stakeholders in this study may not be generalized to other contexts, however, the process and suggestions may be useful when adopting similar interventions in other settings.

6 Conclusion

Caregiver-led and delivered interventions offer an innovative way of supporting caregivers of children with complex disabilities like CP in low-resource settings. The MOC2CTP designed by Malamulele Onward was deemed to be acceptable and needed in a rural setting in Malawi. Several suggestions were also made regarding ways to ensure ease and practical delivery of the intervention in the particular rural context of interest. These aspects of practicality and adaptation are discussed further in a future paper and will be used to improve the design of the program in preparation for its implementation. It is evident that the caregiver training program was found useful and tailored to the needs of the stakeholders. However, the contextual adaptations highlighted are important for the intervention to work optimally. Similar stakeholder involvement is recommended for transferring other innovations which strengthens community ownership and transformative co-development of innovations.

Data availability statement

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

Ethics statement

The studies involving humans were approved by the Human Research Ethics Committee (Medical), University of Witwatersrand, South Africa and the Kamuzu University of Health Sciences Research Ethics Committee, Malawi. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

TB: Conceptualization, Formal analysis, Investigation, Methodology, Project administration, Resources, Writing – original draft, Writing – review & editing. GS: Conceptualization, Methodology, Supervision, Validation, Writing – original draft, Writing – review & editing. WS: Conceptualization, Methodology, Supervision, Validation, Writing – original draft, Writing – review & editing.

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Conflict of interest

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

Publisher's note

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Transferable lessons for care provided to children with intellectual and developmental disabilities based on an analysis of facilitators and barriers to SARS-CoV-2 testing

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Introduction: The purpose of this article is to report on the lessons learned from parents and caregivers of school-age children with intellectual and developmental disabilities (IDD) in Missouri and Maryland regarding the facilitators and barriers to SARS-COV-2 testing.
Methods: Parents participated in interview sessions that employed fuzzy cognitive mapping (FCM), a reliable knowledge-based method that facilitates democratic discourse to understand how stakeholders make decisions. A total of 94 parents from Missouri (58) and Maryland (36) participated in the FCM sessions.
Results: Eight primary barriers and eight primary facilitating factors were identified that influence a successful SARS-COV-2 test. Analyzing the connections between these factors provided valuable information about not only which ideas were most central to the goal of a successful test, but also which factors could be modified to improve the likelihood of success. Results indicate that the physical environment and child preparedness play a central role in successful SARS-COV-2 testing for children with IDD; however, these factors within the context of other invasive procedures should be studied further.
Discussion: It is likely that the findings from this study are transferable to other diagnostic procedures such as influenza, respiratory syncytial virus (RSV), and methicillin-resistant Staphylococcus aureas (MRSA), which require similar testing techniques using a nasopharyngeal swab.

KEYWORDS

SARS-CoV-2 testing, children with IDD, successful treatment models, fuzzy cognitive mapping (FCM), stakeholder engagement

1 Introduction

Recent estimates indicate that almost 18% of children have a developmental disability in the United States, and 1.2% of all US children have an intellectual disability (1). As a group, individuals with IDD have far worse health outcomes than those without IDD (2). For example, people with disabilities report higher rates of chronic conditions than those without disabilities, such as obesity, diabetes, and cardiovascular disease (2). Despite

co-occurring, complex medical conditions, individuals with IDD are less likely to receive preventive healthcare services (3, 4).

There are a variety of barriers that contribute to this disparity in children with IDD. Cognitive and communication difficulties may make it difficult for children with IDD to understand the rationale for medical procedures or to request accommodations from providers (5). Sensory issues and co-occurring psychiatric disorders such as anxiety may also cause distress for children with IDD in healthcare settings (5). Individuals with disabilities also report structural-environmental barriers to healthcare, including ramp unavailability and inaccessible examination rooms and equipment in healthcare settings (6). Process barriers such as a lack of disability knowledge and sensitivity from providers, as well as insufficient time to address one's needs have also been cited by individuals with disabilities (6). The comorbidities associated with IDD and the barriers to healthcare for children with IDD contribute to an ongoing cycle of unmet healthcare needs and risks for this vulnerable population.

Comorbidities and barriers to effective healthcare placed individuals with IDD at a greater risk for severe SARS-COV-2 outcomes compared to those without IDD. Children with IDD are also at a higher risk of SARS-COV-2 infection due to challenges adhering to mitigation strategies. Sensory issues that are common in children with IDD may make it more challenging for them wear a mask. Additionally, social distancing is more difficult for many children with IDD, especially those with complex medical conditions. These children often require assistance with activities of daily living such as eating, during which mask wearing and social distancing cannot occur. In a study of over 64 million patients, people with IDD were more than twice as likely to be hospitalized if diagnosed with SARS-COV-2 than people without IDD (7). Another study found that among children under 17, those with IDD had higher SARS-COV-2 fatality rates (1.6%) than those without IDD (0.1%) (8).

Beyond the direct risks posed by the SARS-CoV-2 virus itself, the pandemic negatively impacted children with IDD and their families in a variety of ways. Children with IDD rely on schools for various services beyond education, including speech, occupational, and physical therapy, social services, and psychological interventions. According to an online survey for caregivers of individuals with IDD, 78% reported that their child lost access to at least one therapy or educational service due to SARS-COV-2 restrictions (9). In addition, Chafouleas and Iovino (10) found that during the pandemic, caregivers of children with disabilities reported significantly higher levels of burden, stress, anxiety, and depression than caregivers of typically developing children. It is abundantly clear that children with IDD are at a high risk for a variety of negative outcomes related to the SARS-COV-2 pandemic.

SARS-CoV-2 testing is a prevention strategy recommended by the Centers for Disease Control and Prevention (CDC) to promote the health and safety of children and their families. Since children with IDDs are disproportionately impacted by SARS-COV-2, access to testing is especially important. However, like other preventive health measures, various barriers interfere with SARS-COV-2 testing for this population. Yet, research surrounding barriers and facilitators to SARS-COV-2 testing for children with IDD is limited. To the authors' knowledge, Haroz and colleagues (11) were the first to examine barriers to SARS-COV-2 testing in a variety of underserved pediatric populations. However, this study was limited to school-based testing and only examined the perspectives of the research teams, rather than key stakeholders such as the caregivers of the children being tested. The current study aims to address this gap in the literature by assessing how caregivers perceive facilitators and barriers to SARS-COV-2 testing for children with IDD, utilizing a participatory methodology called Fuzzy Cognitive Mapping (FCM). Finally, we argue that the lessons learned from this research are transferrable to multiple settings and diagnostic procedures.

2 Method

2.1 Fuzzy cognitive mapping

Fuzzy Cognitive Mapping (FCM) was employed to identify the facilitating factors and barriers to SARS-COV-2 testing among school-age children with IDD. Axelrod (12), a political scientist, was one of the first to use FCM as a means of capturing relational data in a logical and visual format. Specifically, he used it to represent social scientific knowledge using nodes (variable concepts) and edges (causal connections). Later, Kosko and colleagues (13) used FCM to structure virtual worlds (14) and map policy scenarios (15). More recently, social scientists have used FCM to gather the viewpoints that influence decision and sense-making across multiple stakeholders (16–19). These projects have shown FCM to be a reliable knowledge-based model that facilitates democratic discourse to understand how stakeholders make decisions (20). This methodology is highly participatory and fosters social learning between participants. The original purpose for hosting FCM sessions for the present project was to generate community-specific, testable strategies to address social factors that impact the uptake and effectiveness of SARS-COV-2 testing, which will be referred to as "COVID-19 testing" throughout the discussion below.

2.2 Sample

A total of 94 people participated in mapping sessions across Maryland ($n = 36$) and Missouri ($n = 58$). The majority were biological mothers of a child with IDD (74.5%). Participants' ages ranged from 25 to 64 ($M = 44.6$), 89% identified as female, 75% identified as White, 67% were married or partnered, 52% reported working full-time, 82% had post-secondary and higher education, and 22.3% lived in a rural setting (see Table 1).

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TABLE 1 Participant demographic information: overall participants ($N = 94$).

Demographic categories	Missouri		Maryland		Total	
	n	%	*n*	%	*N*	%
Age in years ($M = 44.6$)						
25–30	3	5.2%	0	0.0%	3	3.2%
30–39	15	25.9%	6	16.7%	21	22.3%
40–49	26	44.8%	17	47.2%	43	45.7%
50–59	10	17.2%	9	25.0%	19	20.2%
60–64	2	3.4%	4	11.1%	6	6.4%
Other or missing	2	3.4%	0	0.0	2	2.1%
Gender						
Female	54	93.1%	30	83.3%	84	89%
Male	3	5.2%	6	16.7%	9	9%
Other or missing	1	1.7%	0	0.0%	1	1.0%
Race						
White	47	81.0%	23	63.9%	70	74.5%
Black or African American	6	10.3%	9	25.0%	15	16.0%
Asian	1	1.7%	2	5.6%	3	3.2%
Two or more races	2	3.4%	1	2.8%	3	3.2%
Other or missing	2	3.4%	1	2.8%	3	3.2%
Ethnicity						
Hispanic	2	3.4%	0	0.0%	2	2.1%
Non-Hispanic	55	94.8%	35	97.2%	90	95.7%
Other or missing	1	1.7%	1	2.7%	2	2.1%
Marital status						
Married	27	46.6%	30	83.3%	57	60.6%
Divorced	12	20.7%	3	8.3%	15	16.0%
Separated	2	3.4%	1	2.8%	3	3.2%
Single	7	12.1%	0	0.0%	7	7.4%
Widowed	2	3.4%	0	0.0%	2	2.1%
Living with someone but not married	6	10.3%	0	0.0%	6	6.4%
Other or missing	2	3.4%	2	5.6%	4	4.3%
Employment						
Full time	29	50.0%	20	55.6%	49	52.1%
Part time	7	12.1%	7	19.4%	14	14.9%
Home maker or stay-at-home caregiver	13	22.4%	5	13.9%	18	19.1%
Retired	1	1.7%	2	5.6%	3	3.2%
Unemployed	1	1.7%	2	5.6%	1	1.1%
Other or missing	7	12.1%	20	55.6%	9	9.6%
Highest level of education						
Doctorate degree	3	5.2%	4	11.1%	7	7.4%
Master's degree	14	24.1%	14	38.8%	28	29.8%
Bachelor's degree	16	27.6%	13	36.1%	29	30.9%
Associates degree	10	17.2%	3	8.3%	13	13.8%
Some college	5	8.6%	2	5.5%	7	7.4%
High school/GED	6	10.3%	0	0.0%	6	6.4%
Annual household income						
$100,000–150,000	6	10.3%	9	25.0%	15	16.0%
$75,000–99,999	9	15.5%	3	8.3%	12	12.8%
$50,000–74,999	13	22.4%	1	2.8%	14	14.9%
$20,000–49,999	17	29.3%	2	5.6%	19	20.2%
Less than $20,000	8	13.8%	1	2.8%	9	9.6%
More than $150,000	1	1.7%	14	38.9%	15	16.0%
Other or missing	4	6.9%	6	16.7%	10	10.6%
Setting						
Rural	17	29.3%	4	11.1%	21	22.3%
Urban	9	15.5%	8	22.2%	17	18.1%
Suburban	28	48.3%	22	61.1%	50	53.2%
Other or missing	4	6.9%	2	5.5%	6	6.4%

(*Continued*)

TABLE 1 Continued

Demographic categories	Missouri		Maryland		Total	
	n	%	*n*	%	*N*	%
Parent child relationship						
Biological mother	42	72.4%	28	77.8%	70	74.5%
Biological father	3	5.2%	3	8.3%	6	6.4%
Adoptive mother	7	12.1%	4	11.1%	11	11.7%
Adoptive father	0	0%	1	2.8%	1	1.1%
Other or missing	6	10.3%	0	0.0%	6	6.4%
Number of children with disabilities enrolled in school						
1	41	70.7%	33	91.7%	74	78.7%
2	12	20.7%	2	5.6%	14	14.9%
3	1	1.7%	1	2.8%	2	2.1%
Other or missing	4	6.9%	0	0.0%	4	4.3%

Over 75% of the participants had at least one child with IDD enrolled in school. The participants reported that their children had co-occurring disabilities, with the most common being autism (76%) followed by developmental delay (40.1%), intellectual disability (33.6%), and speech or language impairment (33.5%).

2.2.1 Data collection

The FCM sessions were organized at the community level in partnership with Missouri Family-to-Family and the Kennedy Krieger School Programs. Leadership from partnering organizations worked with the team to recruit parents/guardians who were representative of the gender, ethnic/racial, and social/economic diversity in Maryland and Missouri. Due to the pandemic, FCM sessions took place in an online Zoom format using a program called Draw Chat. This whiteboard program allows collaborators to work on a document in real time via audio and video conferencing systems (e.g., Zoom). Each participant had a blank template on which to draw their maps (see Figure 1). The sessions were capped at 10 participants, but most sessions only included three to six participants.

The research team provided written instructions and a tutorial for all participants. In each FCM session, participants were asked to list up to five important facilitators and five barriers to successful SARS-COV-2 testing among children with IDD using the blank template (Figure 1). Participants then applied directional lines (arrows) depicting the perceived connections between all concepts listed on their maps. Next, they weighted the connections (i.e., 1 = mild connection; 2 = moderate connection; 3 = strong connection) to quantify the relationships between the items on their maps. The research team then facilitated group discussion of the ideas presented on the maps. Conversations during the FCM sessions were audio recorded to provide context to the data on the maps. A minimum of two members of the research team were present for each FCM session.

2.2.2 Data cleaning and preparation

The first step in the data cleaning and preparation process was to conduct an item level analysis of each map (21). Eight members of the research team read each map individually and then developed a codebook (Table 2). This process involved discussing the contents of the maps, identifying code names, and defining codes until a consensus was reached. Ultimately, eight broad facilitator concepts used by the participants and eight barrier concepts were identified.

2.3 Codebook

The same team members then broke into four teams of two researchers. Each team was randomly assigned 24 maps. When a team finished coding their maps, another team coded the same maps without seeing the first team's coding structure. Ultimately, every map was coded independently by each team. The four teams came together weekly to discuss their observations and any questions that arose during the coding process. Finally, the four teams came together to discuss any areas of disagreement on each map until agreement levels reached 100%.

2.3.1 Data analysis

Once the maps were coded, Excel was used to create an adjacency matrix for each map based on the directional and weighted connections identified by the participants. The 94 matrices were then merged into one dataset and exported to other software for final analysis. Specifically, FCMapper, an Excel-based program, was employed to analyze the mapping data using matrix algebra tools of graph theory and to conduct simulation or "scenario" testing. Next, cognitive modeling was conducted using Pajek, a program that operates much like a scientific calculator to visually explore network objects. Using a partitioning feature within this program, variables (or nodes) could be classified within the network by degree of input and output, and subsequently, the graph could be layered to assign nodes with similar values to the same plane. Additionally, by using vectors, the centrality values of each variable could be factored into the overall layout of the model. These centrality values determine the proximity, or "betweenness" of nodes on the model. Finally, structural equation modeling (SEM) was used to confirm the results of the FCM analyses. SEM is a combination of confirmatory factor analysis (CFA) and multiple

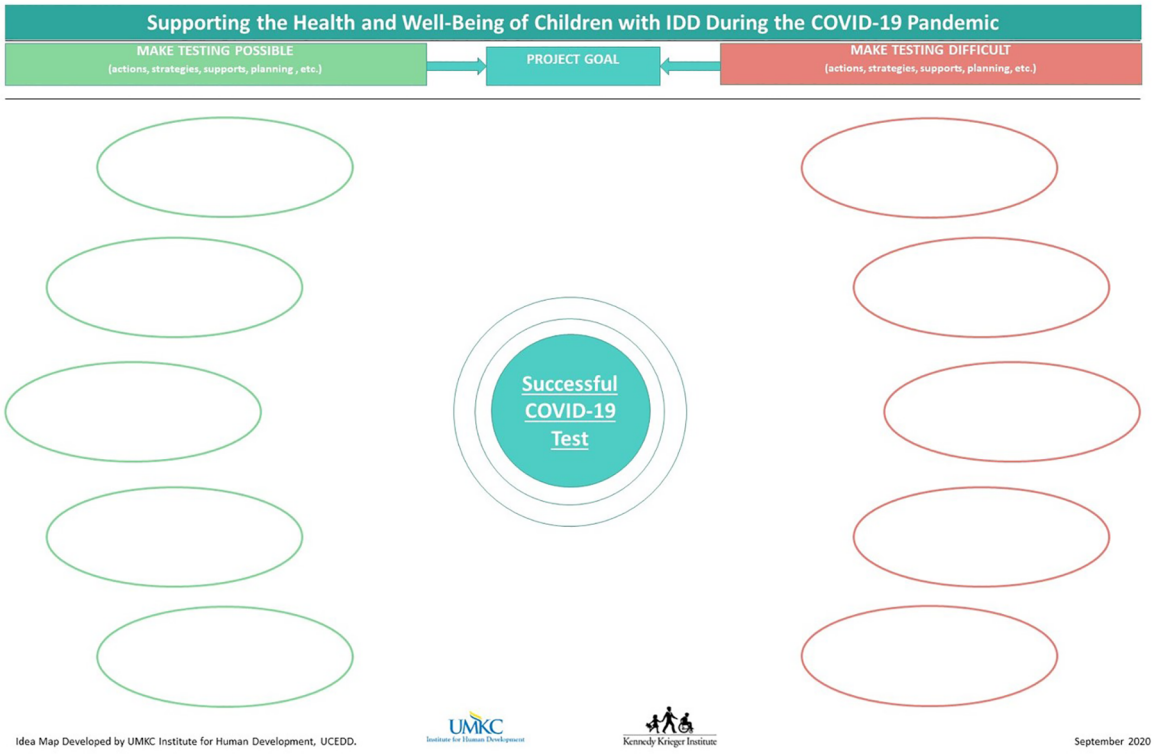

FIGURE 1
Blank fuzzy cognitive mapping template.

TABLE 2 Codebook with definitions.

Codes	Definition
Facilitators	
Preparing the child	Explaining testing process, modeling, social story, practicing, touch testing materials, advance notice, etc.
Parent/guardian involvement	Role the parent wants to play in the testing (i.e., administer the test, restrain, comfort the child)
Good environment	Environment of test is accommodating of different needs, privacy options, number of people, a place the child is familiar with, accessible, familiar support person, etc.
Knowledgeable test administrator	Culturally competent, understanding needs of child with disabilities, compassionate/patient, person-centered practitioner.
Distraction	Bubbles, iPad, fidget toy, comfort item, headphones, etc.
Ease of logistics	Ease of signing up/rescheduling, time of test, testing site, easy to navigate, distance to testing site, etc.
Mode of test (painless)	Swab, saliva, results timing, description of test experience, self-test, etc.
Incentives/motivators/ reinforcers	Reward for taking the test, praise, stickers, food, special toy, etc.
Barriers	
Poor environment	Too much stimuli, bright lights, too many people, wait time, being exposed while testing, safety concerns, lack of supports, unfamiliar place, etc.
Poor test administrator	Tired/grumpy/unprepared healthcare provider, impatient, does not understand unique needs of the child
Costs to family	Financial and emotional costs. Transportation cost, lost wages/time off cost, cost of test, childcare costs for other children, stress, etc.
Child needs	Child needs that can make testing difficult, personality, age, mood, negative feelings about the test, behaviors, etc.
Mode of test (painful/invasive)	Swab, saliva, results timing, description of test experience, self-test, etc.
Inconvenient logistics	Ease of scheduling, distance to testing site, location of test, access to information, etc.
Frequency of test	Having to do the test more than once, negative experience influences the next one, concerns for daily testing, etc.
Stigma	What is said on the news, social media, by peers about COVID19/testing; negative stigma for positive test.

regression (22) that identifies the relationships among latent constructs. For the purpose of the current study, SEM analyses examined the effects of facilitators and barriers related to a Successful SARS-COV-2 Test.

3 Results

3.1 Graph theory

The initial analysis, which employed FCMapper, analyzed the mapping data using matrix algebra tools of graph theory. This allowed the structure of the maps to be examined to determine how participants viewed the actions, strategies, or supports that impact SARS-COV-2 testing. There were 17 variables and 107 connections, and the density index was 0.37. Density is an index of map connectivity, and it is calculated by dividing the number of connections by the maximum number of connections possible between N variables [$D = C/N^2$; (23)]. For the present sample, the total number of possible connections between the 17 variables was 289 connections. Another way to interpret the density score is to recognize that the 107 connections used by our participants only represent 37% of the total possible connections. The participants' maps were not highly complex but they were largely in agreement about how variables impacted a successful SARS-COV-2 test.

Within FCM methodology, there are three types of variables: receiver, transmitter, and ordinary. The variable type is important because it shows how the variables act in relation to one another [(24), p. 51]. Receiver concepts are those that are only affected by other system concepts and have no effect (or output) on other concepts. Participants in the current study were provided with the end point, or goal, for their maps: Successful COVID-19 test. This was the leading receiver variable, as expected, because all other concepts had either direct or indirect influence on the goal. As it turns out, "Successful COVID-19 Test" was the only receiver concept within the data. There were no transmitter concepts resulting from the data, which are variables that only impact others but do not receive input from other variables. Therefore, the remaining 16 variables were ordinary, meaning they played both a transmitting and a receiving function—they both influenced and were influenced by other concepts.

Finally, the centrality of each variable was calculated. Centrality shows how connected a variable is to other variables and the cumulative strength/weights of those connections. In FCM methodology, a variable can be more "central" if the connections to that variable are strong, even though it might have fewer connections altogether (13). As described by Özesmi & Özesmi (24), centrality is "not only a frequency of expression but also how important that variable is given the whole structure of the cognitive map." As expected, the goal (Successful COVID-19 Test) had the highest centrality value (3.28) since it was the ultimate endpoint for the other concepts. The variables with the highest centrality values from there were: 1. Preparing the child (.86); 2. Poor environment (0.65); 3. Good environment (0.64); and 4. Child Needs (0.62). If the two environmental variables are combined, it is apparent that the testing environment- including both positive and negative influences - is highly central to SARS-COV-2 testing among children with IDD. Of note, variables with the lowest centrality values (least impactful to a successful SARS-COV-2 test) were "Costs to the Family" (0.05), "Stigma" (0.06), and "Frequency of Test" (0.11).

3.2 Cognitive modeling

As mentioned above, Pajek was used to visually graph FCM data to display each variable and its connections within the system. Figure 2 shows the cognitive model with all variables (facilitators and barriers) and their connections. The nodes represent the 17 codes used when building the FCM matrix, and the arrows between them represent the directional relationships among these concepts. Facilitating concepts are displayed as squares, and barrier concepts are displayed as triangles. In most cases, participants connected facilitators to the end goal "Successful COVID-19 Test," whereas the barriers had additional connections to one or more facilitators. In these cases, it can be understood that barriers not only inhibited the goal directly, but also inhibited facilitating factors increasing their total negative impact on the goal.

The size of the nodes represents the impact on the overall model, with larger shapes indicating greater impact and smaller shapes indicating weaker impact. The most impactful facilitator, corresponding with the highest centrality, was "Preparing the Child." This concept included ideas such as: providing a social story or video of the procedure; imparting knowledge that the test is coming; ensuring that the child gets a good night's rest before the test; and explaining why the test is being performed. The second most impactful facilitator was "Good Environment," which included ideas such as: minimal waiting times; quieter setting; sensory-friendly environment; and comforting the child. The facilitator concept "Knowledgeable Test Administrator" related to having a calm, flexible person to administer the test; having positive, smiling, and encouraging people to administer; having a patient tester who could slowly explain what will happen as many times as needed; and other factors specific to the person administering the test. Another impactful facilitator was "Mode of Test: Painless," which included ideas such a painless/non invasive test and using a saliva or alternative test instead of a nasal swab.

The most impactful barrier to successful SARS-COV-2 testing for children with developmental disabilities was "Poor Environment." This can be seen near the top of the model on the highest "layer," and it included concepts such as: long wait times; not having available behavioral support; chaotic or busy testing environment; sensory overload problems, etc. The second most impactful barrier was "Child Needs" and included concepts such as: inherent difficulties associated with special needs children (such as mobility); child age and/or challenging personality traits; not having advanced notice about the test; difficulties restraining the child, anxiety, and behavioral meltdowns. Another significant barrier, "Inconvenient Logistics,"

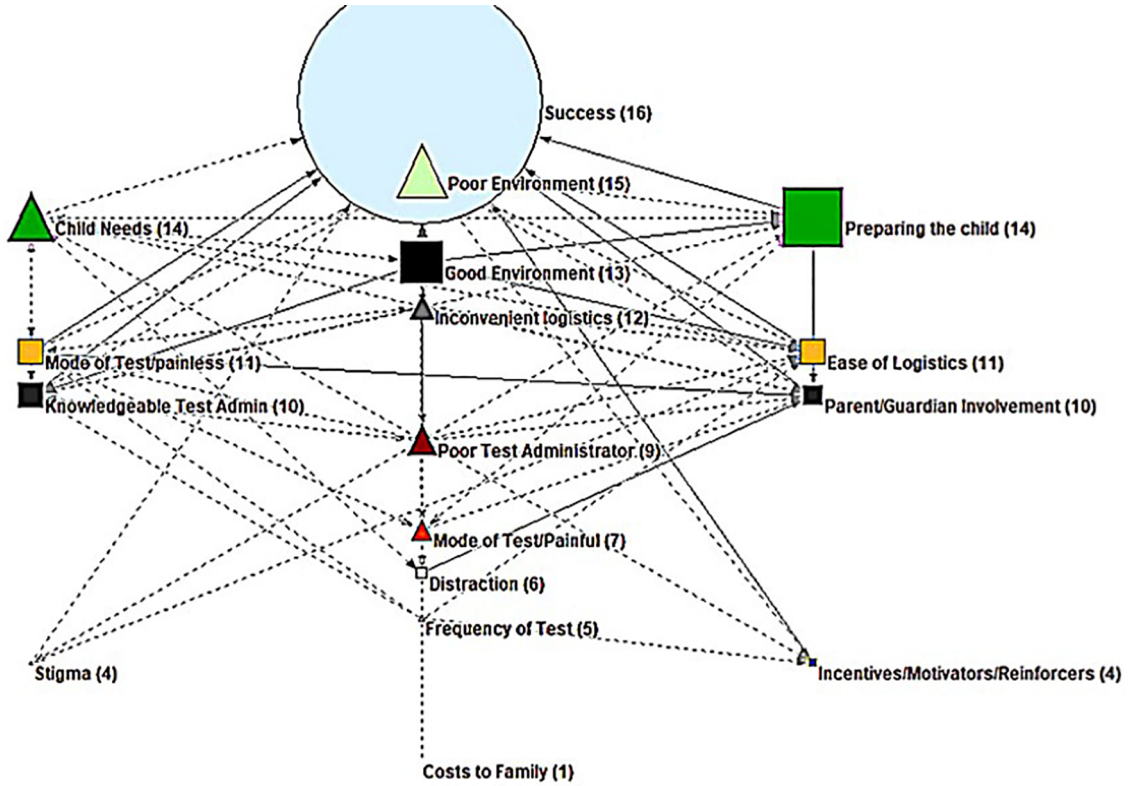

FIGURE 2
Synthesized and modeled FCM.

which included concepts such as distance to the testing site and its location, access to information, and ease of scheduling.

3.3 Structural equation modeling (SEM) results

Finally, structural equation modeling was used to confirm the results of the graph theory and cognitive modeling. There were 16 concepts whose impact on "Successful COVID 19 Test" was estimated. As we mentioned above, eight were facilitators concepts: Preparing the Child ($n = 71$), Good Environment ($n = 61$), Mode of Test: painless ($n = 40$), Knowledgeable Test Administrator ($n = 38$), Ease of Logistics ($n = 38$), Incentive/Motivators ($n = 32$), Distraction ($n = 30$), Parent/Guardian Involvement ($n = 29$). There were also eight barriers: Child Needs ($n = 65$), Poor Environment ($n = 65$), Poor Test Administrator ($n = 53$), Mode of Test: painful/invasive ($n = 44$), Inconvenient Logistics ($n = 37$), Frequency of Test ($n = 12$), Costs to Family ($n = 10$), and Stigma ($n = 5$).

3.3.1 One factor model

In the one-factor model, SEM was used for pinpointing key items that loaded onto Successful SARS-COV-2 Test. Based on the global model fit indices and the factor loadings in each model, variables with the least factor loading (smaller than.3) were removed from the model until acceptable model fit was achieved. Six items remained in the final model. The one-factor model had a good fit: χ^2 (74) = 75.89, $p = .42$, CFI = .97, TLI = .97, RMSEA = .02, SRMR = .06. The path diagram of the structure with the six variables are shown in Figure 3. Results indicated "Preparing the Child," "Good Environment" "Poor Environment," "Poor Test Administrator," "Mode of Test: painful/invasive," and "Child Needs" had a direct effect on Successful SARS-COV-2 Test and thus can be deemed most impactful variables. The SEM results are congruent with the FCM results.

3.3.2 Two-Factor model

In the two-factor model, SEM was used to pinpoint key items that load onto Facilitators and Barriers. Both Facilitators and Barriers were latent variables in this model. The two-factor model had a good fit: χ^2 (73) = 68.72, $p = .62$, CFI = 1.00, TLI = 1.00, RMSEA < .001, SRMR = .06. Results indicated Preparing the Child, Good Environment, Incentives/Motivators have a direct effect on Facilitators (Figure 4). Poor Environment, Poor Test Administrator, Mode of Test: painful/invasive and Child Needs have a direct effect on Barriers.

Table 3 presents the item level estimates and factor loadings below. The one-factor loadings range from .31 to.62, and the two-factor loadings ranged from 0.38 to 0.75 with significant p-values.

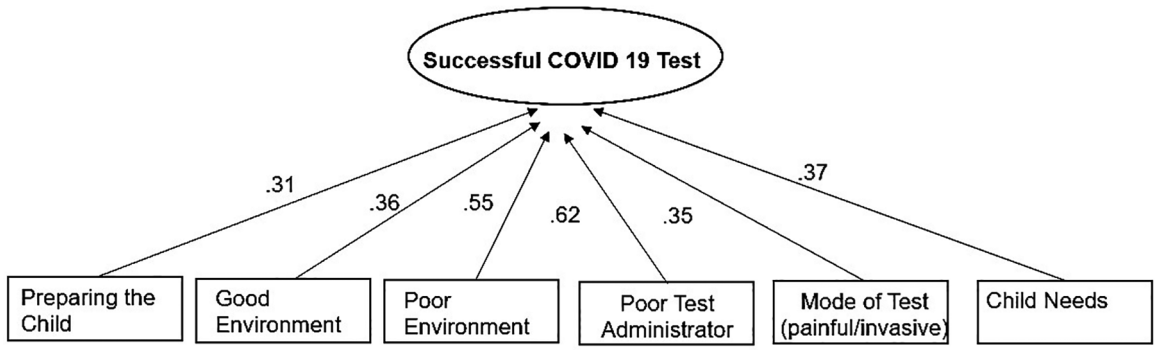

FIGURE 3
One factor SEM model.

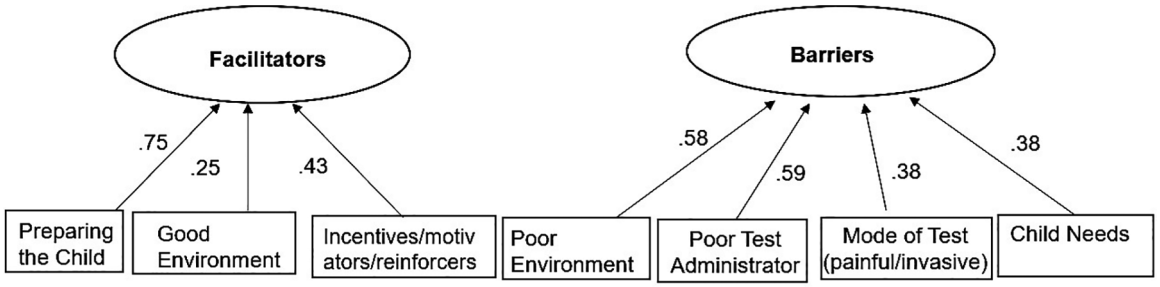

FIGURE 4
Two factor SEM model.

TABLE 3 Factor loadings ($N = 94$).

	One factor loadings		Two factor loadings	
	Effect (λ)	SE	Effect (λ)	SE
Facilitators				
Preparing the child	0.31	0.14	0.75	0.23
Good environment	0.36	0.14	0.25	0.15
Incentives/motivators	-	-	0.43	0.13
Barriers				
Poor environment	0.55	0.14	0.58	0.18
Poor test administrator	0.62	0.14	0.59	0.16
Mode of test (painful/invasive)	0.35	0.14	0.38	0.15
Child needs	0.37	0.14	0.38	0.17

4 Discussion

We used FCM to identify stakeholder-generated, testable strategies for successfully conducting SARS-COV-2 tests among children with IDD. The SARS-COV-2 pandemic presented many challenges and a significant disruption to the routines of daily life, especially for individuals with IDD. Testing children with IDD for SARS-COV-2 carries many considerations that healthcare workers and other test administrators may not be accustomed to. Without specialized training, testing sites may be poorly prepared to handle the physical, behavioral, and other unique needs of children with IDD. This bears with it the risk of unsuccessful testing and potentially misdiagnosed or underdiagnosed cases of SARS-COV-2 in this population. Using the FCM methodology to collect data from families of children with IDD, who are key stakeholders in the testing process, provided valuable information about not only which ideas were most central to the goal of a successful test, but also which factors could be modified to improve the likelihood of success.

4.1 Recommendations

Results showed that the environment plays a central role in the testing experience for children with IDD, including both positive and negative environmental factors. The facilitator "good environment" and the barrier "poor environment" together had the highest centrality in the model. Translating these findings to practice, it is recommended that facilitating SARS-COV-2 testing in children with IDD should initially be aimed at environmental factors. Based on feedback received on the original concept maps reviewed, improving environmental efforts could include trying

to minimize wait times for children with IDD, facilitating a quiet environment, promoting a culture of comfort, and producing sensory-friendly testing areas. Ideas for promoting a sensory-friendly environment might include modifying the color or intensity of light sources, reducing "visual clutter," and offering headphones to children to help reduce ambient noise – all of which are customizable depending on the child's needs (25). Other testing strategies have been developed specifically for students and staff in schools for children with IDD and complex medical conditions. A report by Sherby and colleagues (26) describes three research teams' approaches to testing in various specialized school programs. These teams implemented a variety of techniques, keeping in mind the unique needs of children with IDD. For instance, investigators at Washington University in St. Louis developed a testing procedure that involved collecting a small quantity of saliva, rather than the invasive nasopharyngeal swab procedure utilized by many other SARS-COV-2 testing sites (26). Investigators at the University of Wisconsin-Madison were able to implement in-home testing for school-aged children with complex medical conditions, allowing for parents to conduct the tests on their child (26). Similar to the research described in this article, these projects aim to identify generalizable testing models to increase successful healthcare provision to children with IDD and co-occurring medical conditions.

Preparing the child for the test was also identified as an important facilitator. Ideas for preparing the child, as indicated by the participants, might include providing a social story or video of the procedure, explaining to the child that the test is coming and why it is important, and educating caregivers about the importance of a good night's sleep prior to testing. This finding echoes Haroz and colleagues (11) who recommend identifying and preparing key stakeholders involved in testing. They found that one of the biggest challenges to implementing SARS-COV-2 testing was an inability to engage relevant stakeholders [e.g., families, students, and staff; (11)].

The relationships represented within the FCM model, indicate that making suggested changes may result in improvements in other areas. For example, the FCM model indicates that the "mode of test (painful)" barrier is impacted when environment was altered. Previous evidence suggests that a child who is calm vs. anxious is likely to experience less pain (27). It is also reasonable to expect that a calm, conducive environment will facilitate parent or caregiver involvement, in addition to the confidence and effectiveness of the test administrator.

The barrier "Child Needs" was another central concept within the model. This variable is challenging to address in many ways, since the physical and behavioral needs for children with special needs can vary dramatically. However, the FCM model indicates that increasing child preparedness will lessen the impact of the child's disability-specific needs. This is possibly explained by the extra time spent by test administrators to talk to the child, explain the procedure, allowing them to examine testing implements, which allows test administrators the opportunity to establish a positive social relationship with the child and assess for unique needs. Knowing their needs beforehand and adequately preparing the child, test administrators could better prevent behavioral incidences, reduce anxiety, and avoid restraining procedures. Mitigating child needs through other variables, rather than directly trying to change disability-related needs, is something worth considering.

4.1.1 Transferable lessons

While the scope of this study only models SARS-COV-2 testing in children with IDD, there may be value in extending these recommendations to other testing procedures within this population. Results indicate that environment and child preparedness play a central role in successful SARS-COV-2 testing for children with IDD; however, these factors within the context of other invasive procedures should be studied further as it is likely that the findings from this study are transferable to other diagnostic procedures. For example, many other infections such as influenza, respiratory syncytial virus (RSV), and methicillin-resistant Staphylococcus aureas (MRSA) require similar testing techniques using a nasopharyngeal swab. Testing for Strepotococcus (strep throat) involves a throat swab, which may be a similarly distressing experience to children with IDD. Opportunities to improve procedural experiences for children with IDD could even be extended to areas such as: needle sticks for vaccines, medication administration, and blood draws, blood pressure checks and application of monitoring devices, care and dressing of wounds, dental and vision procedures, fitting for orthopedic devices, and others. It is likely that improving child preparedness and the environmental space in which these procedures are performed could have far-reaching impact within a variety of settings, including medical offices, school nursing offices, dentistry, therapy centers, stand-alone testing sites, and even home-based health services. Thus, it is worth considering how these recommendations could be applied in the daily practice of any healthcare professional, including therapists, dentistry specialists, school nurses, nursing assistants, laboratory and imaging personnel, and even volunteers.

Altering the environment and taking the time to prepare a child may seem like daunting tasks in fast-paced healthcare settings that already struggle to manage patient loads. If so, it may be beneficial for clinical leaders to help staff identify and start with small, manageable changes, such as dimming light sources or turning on calming music as well as allowing a known caregiver to attend. Additionally, it is helpful to remember that the extra time it might take to prepare a child for a procedure is not only important for that child's well-being, but it can also prevent challenging and time-consuming situations resulting from behavioral crises.

4.2 Limitations

While FCM methodology provides an organized, structured process for analyzing qualitative data, there is the potential for bias to be introduced while coding qualitative data for emerging themes. Steps were taken to minimize this bias by dividing cognitive maps between teams, coding them separately, and then meeting together to discuss coding results and areas

of disagreement. Additionally, although strong correlations were identified between certain concepts and the goal of a successful SARS-COV-2 test, this study did not specifically evaluate different interventions that could enhance these concepts. The impact of specific environmental changes on children with IDD within the testing area, as one example, should be studied further.

The race and ethnicity of the samples in both Missouri and Maryland were not fully representative of the state populations in 2021, when data were collected. For example, in Missouri, White residents were 77% of the state population but they made up 81% of the Missouri sample. Black or African American participants (10.3% sample, 11.4% state) and Hispanic participants (3.4% sample, 4.9% state) were slightly underrepresented (28). In Maryland, 25% of the sample identified as Black or African American, slightly under the statewide population statistics (29.5%). Hispanic residents were not represented in the Maryland sample even though 11.8% of Maryland's population was Hispanic. The percentage of White study participants in Maryland (63.9%) was over representative of the statewide population [48.7%; (29)]. Other races and ethnicities, including Asian and American Indian and Alaska Natives, were underrepresented in both states.

5 Conclusions

As impacts from the SARS-COV-2 pandemic linger, new variants of the virus emerge, and new infection control recommendations are developed, testing children with IDD needs to be a viable option for proper diagnosis and treatment. Children with IDD are especially vulnerable to the effects of the pandemic, and this population brings many unique needs that may be challenging for testing sites to manage. The FCM process helped evaluate both facilitating and inhibiting variables surrounding successful SARS-COV-2 testing from the perspective of key stakeholders. This provided valuable insight into which of these variables are most central to success and where efforts can be applied to leverage the highest improvement potential.

The FCM model revealed that environmental factors, preparing the child for the test, and child needs were the most central concepts surrounding successful testing. Other important variables included logistics, mode of test (painless), knowledgeable test administrators, and parent/guardian involvement. Costs to family, stigma, and frequency of the test were least central. Through scenario testing, it could be seen that environmental changes had a positive influence on preparing the child for the test as well as the goal of a successful test. Additionally, improving child preparedness by itself had a positive impact on the overall model. The best result obtained through scenario testing, however, was when environment and child preparedness were improved simultaneously. This indicates that to achieve a successful SARS-COV-2 test in children with IDD, efforts should be focused on both environmental modifications as well as steps to prepare the child for the test.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Washington University in St. Louis Human Research Protection Office. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided audio recorded verbal consent to participate in this study.

Author contributions

GG: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Validation, Writing – original draft, Writing – review & editing. JB: Data curation, Formal Analysis, Visualization, Writing – original draft. RN: Writing – original draft. CE: Writing – original draft. DC: Formal Analysis, Validation, Visualization, Writing – review & editing. MS: Funding acquisition, Project administration, Writing – review & editing. EJ: Data curation, Formal Analysis, Writing – review & editing. MM: Data curation, Formal Analysis, Writing – review & editing. LK: Conceptualization, Funding acquisition, Supervision, Writing – review & editing.

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Conflict of interest

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

Publisher's note

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Project SCOPE: a National Training Initiative to improve capacity of providers to support children impacted by prenatal opioid exposure using the ECHO model™

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Introduction: Neonatal abstinence syndrome (NAS) is a complex condition resulting from prenatal substance exposure that has become increasingly prevalent as a result of the opioid epidemic. NAS can lead to long-term developmental challenges. Interdisciplinary teams with experience working with children with disabilities that focus on social determinants of health can be effective at supporting families affected by NAS. Unfortunately, interdisciplinary teams often lack sufficient training, ongoing practice support, and public health policies to support these families. The objective of this project was to determine the feasibility and impact of a National Training Initiative, called Project SCOPE, to improve the capacity of providers to address the needs of children with NAS and their families.

Methods: Fourteen (14) sites were trained to fidelity in the ECHO model and SCOPE curriculum, and then each team implemented this model for at least one, eight to 12 session series between 2019–2022. The reach, impact, satisfaction, and intention to implement the model were assessed from administrative records, pre/post surveys, and post-session evaluations.

Results: SCOPE state teams delivered the curriculum to 9,392 individuals across 33 US states. Surveys from 2,197 individuals were used for analysis. Most participants (84%) had previous training in trauma informed care, but only 53% had any training on the NAS or the opioid crisis' impact on children. Satisfaction with SCOPE sessions was high (96.4%), and there was a statistically significant increase of self-reported knowledge change from pre- to post-SCOPE. Over 94% reported their skills increased because of participation. Over 97% of participants indicated their motivation to work with this population increased from SCOPE participation and that they could successfully apply what they learned. Almost 70% reported they were "very" or "extremely" likely to use their new skills.

Discussion: Project SCOPE is a highly effective and impactful model that can radically improve capacity to support children affected by the opioid epidemic, thereby increasing the capacity of our healthcare system to respond to this epidemic. Moreover, this model can be rapidly deployed and

reach a wide geographic region, especially areas that are affected by the opioid crisis and underserved rural communities.

KEYWORDS

Neonatal Abstinence Syndrome, Neonatal Opioid Withdrawal Syndrome, early childhood, interprofessional education, ECHO

Project SCOPE: a National Training Initiative to improve capacity of providers to support children impacted by prenatal opioid exposure using the ECHO model™

Substance use disorders (SUD) during pregnancy can lead to Neonatal Abstinence Syndrome (NAS)[1] in prenatally exposed children (1). Similar to those with developmental delays or disabilities, children with NAS may present with complex physical, mental, and behavioral health and social support needs requiring the collaboration of experienced practitioners, researchers, and leaders across domains of healthcare, public health, early intervention, education, mental and behavioral health, family support, and social services (2, 3, 46). Therefore, caring for and educating children with a history of NAS presents unique challenges given the cross-system collaboration necessary to effectively reduce the public health burden.

Unfortunately, the existing healthcare workforce is insufficient to address the clinical needs of the growing population of infants with NAS or mitigate the public health impact. For example, since 2010, Maternal Opioid-Related Diagnoses (MOUD) almost doubled from 4.6 per 1,000 delivery hospitalizations to 8.2 per 1,000 (4). The increase in opioid use during pregnancy has contributed to an increased U.S. prevalence of NAS from 4.0 in 1,000 births in 2010 to 7.3 in 1,000 births at the peak in 2017 (5). Infants with NAS are at increased risk for a variety of complications shortly after birth, such as tremors, seizures, feeding problems, and they may require pharmacological treatment and/or intensive care admissions (6). Clinical care research has emerged to demonstrate effectiveness of the Eat, Sleep, Console approach to decrease length of hospital stay compared to usual care (48). However, while most infants recover from the immediate withdraw symptoms (7), there is growing literature to suggest that NAS can have effects that persist well into early childhood and adolescence (8–13). For instance, children between the ages of 3 and 8 years with a history of NAS are more likely to be diagnosed with developmental delay or speech impairment and more likely to receive school-based supports (8). Preschool-aged children with a history of NAS have also shown deficits in motor skills and memory ability, poor peer relationships, hyperactivity and short attention span, impulsivity, aggressive behavior, and increased risk of developing Attention-Deficit/Hyperactivity Disorder [ADHD (9, 10, 12–17)].

Given the possible multi-faceted needs of children with NAS and number of systems that may be required to effectively support these children, interdisciplinary team-based approaches are apt to be more effective (18). Moreover, there is growing recognition that the conditions of the environments where people are born, live, learn, work, play, worship, and age affect a wide range of health, functioning, and quality-of-life outcomes and risks (US Department of Health and Human Services). Addressing these social determinants of health [SDOH (19)] requires that a robust interdisciplinary workforce has access to training and continuing professional development on emerging evidence-based SDOH practices.

Indeed, public health approaches [e.g., the 10 essential public health services (20)] call for equitable access for all by building a diverse and skilled workforce, based on innovative, evidence-based programs that ensure strong organizational infrastructure for public health. Therefore, innovative training approaches/models are needed to enhance the capacity of interdisciplinary teams. However, to ensure effectiveness of such programming, the training delivery method should be flexible, collaborative, acceptable, and effective. Such programs should have minimal lead time from conceptualization to implementation to ensure rapid uptake of best-and practices and should easily adapted for use by various communities to meet their own unique needs. One such promising delivery method is the ECHO model™ (21).

The Extension of Community Healthcare Outcomes model for professional development

The Extension of Community Healthcare Outcomes, or Project ECHO, was developed as a model of professional development (PD) that adheres to the principles of effective PD (21–25). Specifically, content delivered through ECHO is directly relevant to current professional struggles (26, 27) and includes four core features of effective adult learning: (1) delivery of content knowledge, (2) active learning (i.e., hands on work), (3) integration of changes into daily professional life, and (4) continued follow up and support after training to allow for continued adaptation of the skills (24, 27–31).

The Project ECHO model includes (1) short professional development trainings, (2) case-based learning through case-narratives, (3) ongoing co-management and individual coaching and (4) robust program evaluation (21). All of this is conducted over video conferencing, which allows professionals in distinct settings to receive highly relevant PD and receive specific guidance related to their current needs and concerns. This means that interdisciplinary teams are trained to work more effectively with

1 Neonatal Opioid Withdrawal Syndrome (NOWS) has been the term used to describe specific prenatal opioid exposure in infants (47); however, we will use the more inclusive term NAS throughout this paper.

their clients, rather than simply be presented with general and vague information that may not be relevant to their current situation. Participants of ECHO are, therefore, able to be better prepared to respond to emerging public health needs, which improves the capacity and diversity of the workforce that supports children with NAS. Providing ongoing support to those working with children and families impacted by the opioid crisis is consistent with The 10 Essential Public Health Services (20).

The primary goal of the current project was to develop, disseminate, and evaluate the implementation of a model of PD using a core curriculum to meet the immediate and complex needs of the workforce supporting children and families impacted by the opioid crisis, specifically children with a history of NAS. Project SCOPE (Supporting Children impacted by the OPioid Epidemic) aimed to enhance the public health workforce rapidly and effectively to increase capacity for interdisciplinary community providers to address this issue. Project SCOPE was a 3-year National Training Initiative (NTI) intended to build nationwide provider capacity and confidence in applying evidence-based practices. The primary goal was to determine the impact of a large, multi-state implementation of Project SCOPE and its potential to build healthcare workforce capacity to address NAS on a large scale. This project was based on the ECHO model to ensure that it can quickly and effectively meet the needs of a broad range of communities that are impacted by public health issues related to NAS and the opioid crisis.

Method

Project SCOPE National Training Initiative

Project SCOPE was a collaboration between three University Centers of Excellence in Developmental Disabilities [UCEDD; i.e., Wyoming Institute on Disabilities (WIND), Nisonger Center, and University of Cincinnati Center for Excellence in Developmental Disabilities (UCCEDD)]. The UCCEDD and Nisonger Center developed the SCOPE curriculum, and WIND leveraged their expertise with the ECHO training model to structure the implementation of this curriculum. The NTI lead team trained 14 state teams to implement the core SCOPE curriculum developed during a 2018–2019 pilot phase. During the spring of 2019, a robust evaluation plan was implemented for the pilot study to determine the feasibility, reach and impact of this model of PD. Four key outcomes were evaluated: (1) network reach and participation, (2) participant satisfaction and relevance to their practice, (3) participant knowledge and skill development, and (4) participant intention to implement new skills. This framework was then replicated for the current NTI project. A base set of evaluation measures created during our pilot phase was used at all sites; although, each site could add additional measures as desired. Note that additional site-specific measures are not reported here. During the fall 2019 to spring 2020, state teams applied to receive training on the ECHO model and core SCOPE curriculum. Each state team was led by a UCEDD and/or Leadership Education in Neurodevelopmental Disabilities (LEND) program. UCEDDs and LENDs are part of a nationwide network of centers that are enabled by the Developmental Disabilities Rights Act (32) to provide training, research, supports, model programs and other supports to address the needs of those with disabilities.

The following states implemented SCOPE as part of this NTI project: Arizona, Colorado, Georgia, Kentucky, Maine, Minnesota, New Hampshire, New York, North Dakota, Ohio, South Dakota, Utah, Vermont, and West Virginia. Maine and New Hampshire served as a joint team for this project. Each state was trained to fidelity in the use of the ECHO model and the SCOPE curriculum. This was done remotely given COVID-19 restrictions. Two-day training sessions were conducted by WIND ECHO Superhub staff (experts in the ECHO model) and content experts on NAS from the Nisonger Center and UCCEDD. The training team included clinical psychologists, public health professionals, occupational therapists, and other professionals with NAS expertise. Each state team then implemented SCOPE and was responsible for recruitment, delivery and evaluation of their state implementation using a standardized evaluation framework developed by the lead sites. Each site adapted the implementation timeline to meet their local needs, which ranged from 8 to 12 weeks and was implemented between 2019 and 2022. Given that ECHO is delivered over Zoom™, the public health emergency due to the COVID-19 pandemic did not impact implementation significantly. However, the use of teleconferencing as a primary means of interaction was still new to some of the NTI's target professionals at this time. All data were collected electronically using REDCap (33) and iECHO (a proprietary administrative reporting tool used by all ECHO implementation sites). Each site received $17,500 for participation in the training and implementation of three SCOPE series.

Participants

After each state team was trained, they implemented SCOPE locally. Practicing professionals (N = 9,392) were recruited by the local state teams through a variety of methods such word-of-mouth, e-mail, and social media. Programs that were targeted for recruitment included state-wide early intervention systems (Individuals with Disabilities Education Act Part C), early childcare and education (e.g., preschool, childcare, and Head Start centers), healthcare professionals serving children birth to age five, and family members. Participants were free to join Project SCOPE sessions at no cost. Each implementation site was free to use an open or closed cohort design. Participants were eligible if they worked with children at risk for NAS, regardless of professional capacity, or were family members of children with NAS. All participants were invited to complete all surveys by the host site, which allowed them to register to participate in the SCOPE sessions.

Measures

Evaluation of SCOPE focused on the following outcomes: (1) network reach and participation, (2) participant satisfaction and relevance to their practice, (3) participant knowledge and skill development, and (4) participant intention to implement new skills.

Data sources included administrative measures, traditional pre-post surveys at the beginning and end of SCOPE participation, and short evaluations after individual ECHO session. For pre-post surveys and session evaluations, a retrospective pre- then post-survey was used to account for reporter bias (34). Specific measures for each of our four outcome areas are below. Note that participation was completely voluntary, and participants were free to skip any question.

Network reach and participation

These data came from three sources: administrative data from the iECHO platform, a 25-item demographic form, and a survey of to what degree adverse childhood experiences impact their clients. The iECHO platform is the standard administrative data reporting tool used by all ECHO implementation sites. It captures the name, contact information, and practice location of all attendees. Name and contact information were redacted for this analysis. Zip code was used as a proxy for participant location given that many participants joined from extremely remote locations. Zip codes were converted to county to limit the potential for inadvertent identification of participants in extremely rural or frontier areas. These data were then used to generate heat maps by state and county to understand the reach of SCOPE.

A 25-item demographic form captured information on participant age, race, ethnicity, income, education, job, job setting, years of job experience, years of childcare experience, experience with NAS and trauma related care, and interest in topics related to NAS. This form was internally generated by the research team. The adverse childhood experiences survey is an internally generated 22-item questionnaire asked participants to indicate "Yes" or "No" if they worked with any children who had experienced or were experiencing any of the listed adverse experiences. If a participant responded "Yes" to a particular experience, they were asked to indicate the number of children they were working with who had that experience. Both these instruments were used to characterize the SCOPE participants, as well as the degree to which their clients have experienced adverse experiences using descriptive statistics.

Participant satisfaction and relevance to their practice

Participant satisfaction and relevance to their practice was captured using the post-session evaluations and the final post-SCOPE survey at the end of an entire series. Satisfaction with the speakers and topic(s), relevance to their work, and usefulness of the material were captured following each session. The post-SCOPE survey asked questions about an individual's overall learning and experience during their participation, evaluating improvement of one's quality of practice, further demonstrating session relevance.

Participant knowledge and skill development

Knowledge and skill development were evaluated using an internally generated opioid knowledge survey (OKS), post-session surveys, and the post-SCOPE survey. The OKS was an objective measure of knowledge of the learning objectives covered as part of SCOPE sessions. Participants completed the OKS before attending any SCOPE sessions and after the SCOPE series had concluded. The first version of the OKS contained 15 multiple-answer and multiple-choice items and scores ranged from 0 to 15, where participants received a point for each correct answer. The OKS was revised in 2021 to add six additional items to further explore knowledge gain. Both versions were scored using a sum score and change from pre- to post- using paired sample *t*-tests.

Post-session surveys identified participants' self-reported knowledge before and after the session, as well as the likelihood of using the knowledge they had gained. Post-SCOPE surveys identified knowledge increase, skills increase, likelihood of continuing to use new knowledge/skills, and a retrospective pre-post measure of knowledge related to learning objectives prior to the session, current level of knowledge, and participant rated level of knowledge needed to be successful in a participant's job. All items were on 5-point Likert scales.

Participant intention to implement new skills

The intention to implement new skills was captured in both post-session and post-SCOPE surveys. For post-session surveys, intention to use new skills was measured via a 7-point scale ranging from 1–False to 7–True. After each session, participants were asked to endorse items that framed the strategies and skills as: "I plan to…," with session strategies and skills listed at the end of each item. Confidence in one's ability to implement new information or skills, plans to use new skills, how often participants predicted using skills, and a timeline to implementation were evaluated in the post-SCOPE survey. Participants also indicated whether they had used specific skills and if they intended to use those skills in the future. Additionally, participants were asked to report whether their quality of practice and motivation had increased due to attending SCOPE sessions.

Procedure

After each state team was trained, they implemented SCOPE at least once. Prior to implementation, each site executed a licensing agreement with the ECHO Institute to implement ECHO to fidelity. Each SCOPE series ran between eight to 12 sessions. Aligned with the ECHO model, the exact number of sessions was decided upon by the state teams based on local needs. All SCOPE sessions met on a consistent schedule for either 60 or 90 min each at a time convenient to accommodate professional schedules. Sessions followed a specific structure to maximize learning, engagement, and application of best practices. Sessions included introductions and a short didactic presentation by a content expert followed by a case-based learning opportunity.

All evaluations were administered using a REDCap™ (33) database managed by the University of Wyoming. All state lead team sites collected data related to the sessions they conducted. Separate REDCap™ projects were created for each site. Sites were allowed to add additional questions to a predetermined core evaluation; however, no questions were allowed to be removed from the core evaluation. Participants completed measures through a REDCap™ database prior to their first SCOPE session (pre-SCOPE) and following each session attended (post-session) as

well as following the entire series (post-SCOPE). Demographic and adverse childhood experiences forms were completed pre-SCOPE. The OKS was collected pre- and post-SCOPE. The post-SCOPE survey was completed after the conclusion of each sites' SCOPE series.

Analysis

This is a program evaluation of an NTI using the ECHO model to improve workforce capacity. We used descriptive analyses to explore each of the outcomes of interest listed above. Means, standard deviations, percentages, heatmaps, and bar charts were used to describe the distributions and visualize the data. Where appropriate, changes from pre- to post-survey were tested using paired sample *t*-tests (alpha set at 0.05) and descriptive statistics. All analyses were conducted using SPSS version 28.

To examine the reach of SCOPE, we created heatmaps to visualize the location of participants, along with density of participation. We also characterized the number and types of practitioners that were trained through this model, as well as the estimated caseload of children potentially impacted by the training. We then calculated the average satisfaction of all sessions, across all sites. Next, we evaluated the impact of the training on the core knowledge and skills of the providers. Subjective changes in knowledge were measured during pre- and post-SCOPE surveys and surveys following each session. This was captured using a traditional pre-post design, as well as a retrospective pre-post to account for bias. These were then compared to the respondents' knowledge goal (i.e., what level of knowledge was desired). Also, paired sample *t*-tests were used to measure knowledge changes after each session. Note that in the case of the OKS, separate paired sample *t*-tests were used for each version. Finally, we evaluated the providers' intention to implement their newly acquired skills, their use of the strategies or skills, and implementation of the knowledge gained as a part of the SCOPE community. Means, standard deviations, and bar charts were used to characterize these measures.

Results

Characteristics of participants

Based on administrative data from iECHO, 9,392 individuals participated in at least one ECHO session and came from 33 states: Arizona, California, Colorado, Connecticut, Florida, Georgia, Idaho, Indiana, Iowa, Kansas, Kentucky, Maine, Maryland, Massachusetts, Michigan, Minnesota, Montana, Nebraska, New Hampshire, New York, North Carolina, North Dakota, Ohio, Oklahoma, Oregon, South Dakota, Texas, Utah, Vermont, Virginia, West Virginia, Wisconsin, and Wyoming. Figure 1 shows a heatmap of the US with state and county as the units of analysis. As expected, most participants came from states that had teams participating in this project. However, many participants joined from all over the country. Density of participation varied greatly from state to state. Given that many states are considerably more rural than others, that is not unexpected. However, many states had broad participation from a majority of their counties.

A total of 2,197 individuals provided at least some demographic or professional background information. Each question was analyzed separately to maximize the sample, which varied between 1,968 and 2,172 respondents. The overall sample represented a wide range of backgrounds and professions. Those identifying as women accounted for most participants. Men and those not reporting a binary gender accounted for the minority of participants. Most participants were highly educated with 89.9% possessing a bachelor's, graduate, or professional degree. Most participants identified as White, but there were significant proportions from other racial and ethnic groups. Black or African American and Hispanic/Latino were the largest non-White groups, with all other groups accounting for small percentages. Age was normally distributed, with most participants between 35 and 54 years old. See Table 1 for demographics.

Participants' occupations were diverse. The most commonly reported occupation was "other," ($n = 752$) although, participants included social workers ($n = 298$), early intervention specialists ($n = 214$), education professionals ($n = 63$), medical professionals (Physicians, developmental behavioral pediatricians, child psychiatrists, nurse practitioners, $n = 61$), legal professional (attorneys, judges, guardian ad litems; $n = 7$). The average years in current professional role was 8.29 years (SD = 8.41), but there was a large range (from 0 to 47 years). The average number of years working with children aged 0–5 was 12.74 years (SD = 10.37), ranging from 0 to 50 years of experience. The current work setting of the participants was predominantly home-based, which is consistent with the age of the children who would be affected by this project. However, there were many participants from other settings such as schools, clinics, and childcare organizations. The participants served in a wide range of systems, ranging from government (e.g., Part C), hospitals and clinics, and childcare programs. However, the majority worked in "Other," which were not one of the predetermined answer choices (see Table 2).

Most participants reported working with children who had been exposed to opioids *in utero*. The average number of children on their caseloads with opioid exposure was 5.6 children, accounting for 141,085 children across all training sites, indicating a substantial impact of opioid exposure which is consistent with the ongoing opioid crisis in the US. Most participants (84%) had received some training in trauma informed care, but only 53% had received training on the opioid crisis and its impact on children, suggesting limited training option on the impact of NAS.

Satisfaction and relevance

Satisfaction with SCOPE was high, with 96.4% of participants reporting satisfaction with the sessions and 96.1% endorsed that the sessions contributed to their overall understanding of strategies to support children impacted by NAS. Additionally, in relation to personal relevance, 98.1% of participants felt that the learning topics were relevant and 96.1% of participants reported the weekly sessions contributed to their understanding of the opioid crisis and how it impacts children (see Figure 2). Further, most participants felt attending the sessions helped them

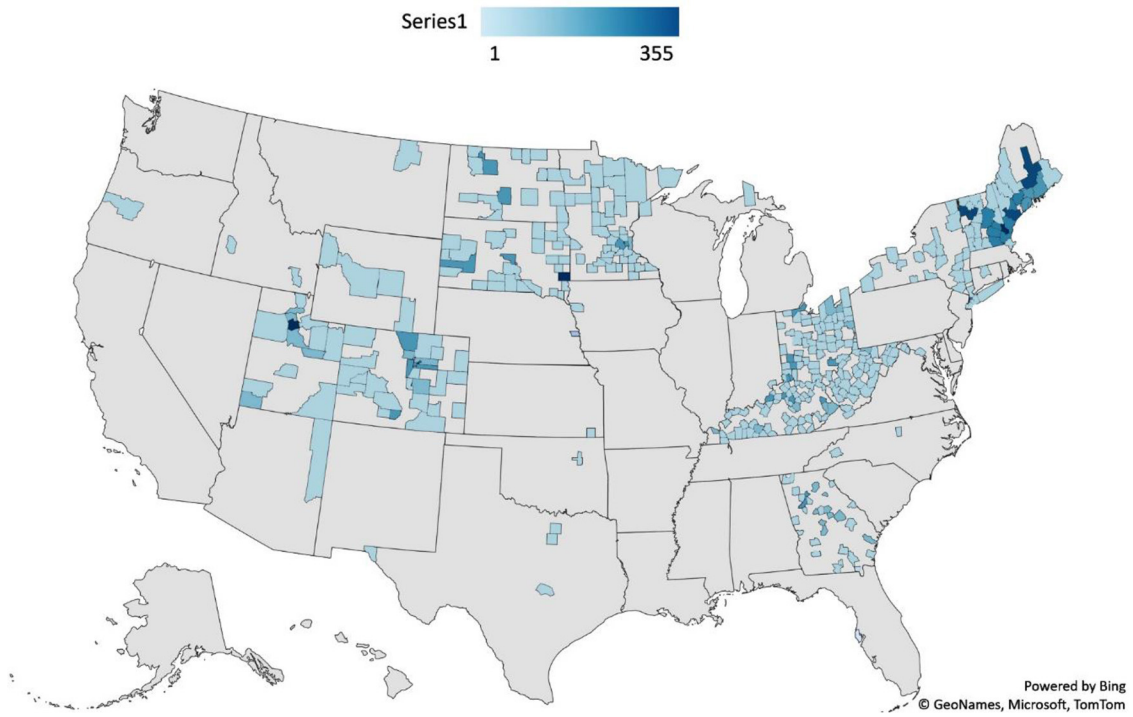

FIGURE 1
Location of participants in SCOPE by state and county.

connect to other practitioners, and that SCOPE expanded their professional networks.

Knowledge and skill gains

Opioid knowledge survey results

Results for version 1 of the OKS (prior to 2021) showed a statistically significant increase in knowledge from pre (M = 8.50, SD = 1.65) to post (M = 9.02, SD = 1.40); $t_{(114)} = 3.33$, $p = 0.001$. The effect size, measured by Cohen's *d*, was $d = 0.31$, indicating a medium to small effect. Similarly, results for version 2 showed a statistically significant increase in knowledge from pre (M = 13.07, SD = 2.13) to post (M = 14.02, SD = 2.04); $t_{(55)} = 2.54$, $p = 0.007$. The effect size was also medium to small with a Cohen's *d* of 0.34.

Post-SCOPE survey results

Most participants (96.1%) rated knowledge improvement due to participation in SCOPE sessions, and 94.5% felt that their skills improved at least some. There was a significant increase in self-reported knowledge from before participating in SCOPE (M = 3.32, SD = 1.0) to after participating (M = 4.17, SD = 0.94), $t_{(410)} = 23.43$, $p < 0.001$ (see Figure 3). The effect size was large, with a Cohen's *d* of 1.16. Though, the knowledge level after participating in SCOPE (M = 4.17, SD = 0.94) was not significantly different than the knowledge level participants reported they need to be successful (M = 4.12, SD = 1.07), $t_{(411)} = 1.17$, $p = 0.24$. The effect size was small with a Cohen's *d* of 0.06. Knowledge also significantly increased following each of the sessions. Using a retrospective pre-then post design, respondents reported a significant increase in knowledge from before the training (M = 2.74, SD = 0.74) to after the session (M = 3.51, SD = 0.60), $t_{(1,560)} = 54.07$, $p < 0.001$. The effect size was large with a Cohen's *d* of 1.37.

Intention to implement

Ninety-two percent reported their practice quality improved due to participating in Project SCOPE. Further, 97.4% of respondents said that their motivation to work with children impacted by opioids had increased. Importantly, 97.8% of participants, or 5,072 of the 5,186 responses to weekly session surveys, said that they could successfully apply what they learned in their practice. This confidence is confirmed from their actual usage, with 84.0% of respondents indicating that they had used new knowledge from the SCOPE session in their work. Moreover, it seems once trained, participants could rapidly implement what they had learned. When asked the degree of how likely they would use their new knowledge, 93.1% said they were at least moderately likely to use their new knowledge, with 69.0% saying they were "very likely" or "extremely likely" (see Figure 3).

Participants were asked to report on their confidence related to the use of specific strategies or skills taught in the SCOPE sessions and their intent to use these strategies and skills in the future. Fifty-six percent of participants reported having used the strategies at the time of the post-SCOPE survey. Further, 67.0% said that they intended to use one more of their new skills in the future. Moreover,

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TABLE 1 Demographic characteristics of SCOPE participants.

	n	%
Participant location**		
Colorado	1,153	12.4
Georgia	822	8.8
Kentucky	1,068	11.4
Maine	702	7.5
Minnesota	436	4.6
New Hampshire	872	9.3
New York	572	6.1
North Dakota	199	2.1
Ohio	1,278	13.6
South Dakota	747	8.0
Utah	673	7.2
Vermont	508	5.4
West Virginia	202	2.2
Other	160	1.7
Total	9,392	
Gender		
Female	2,052	94.6
Male	111	5.1
Other/not reported	5	0.2
Total	2,168	
Education level		
Some High School	1	0.0005
High School Graduate or GED	30	1.4
Some College or Post-High School or 2 Year Degree	189	8.6
College Graduate	832	37.9
Advanced Graduate or Professional Degree	1,120	51.0
Total	2,172	
Race		
White	1,781	82.6
Black	163	7.6
Asian	29	1.3
Hispanic or Latino	86	4.0
Prefer to Self-Describe	94	4.4
More than 1	2	0.1
Total	2,155	
Age		
<18	15	.7
18–24	78	3.6
25–34	467	21.3
35–44	570	26.3
45–54	601	27.7
55–64	342	15.8
65+	100	4.6
Total	2,167	

**Data come from iECHO system, states implementing SCOPE listed with all other states listed as "other."

TABLE 2 Professional experience of SCOPE participants.

	n	%
Primary system of profession		
Part C/Early Intervention	464	21.9
Part B/Special Education	114	5.4
Head Start	157	7.4
Early Head Start	57	2.7
Childcare	153	7.2
Home Visiting	197	9.3
Hospital/Medical Center	230	10.8
Private Practice	54	2.5
Other	696	32.8
Experience working with children with prenatal opioid exposure		
Yes	1,365	64.2
No	478	22.5
I Don't Know	282	13.3
Received any other training related to trauma or trauma-informed care		
Yes	1,796	83.8
No	347	16.2
Received any other training on the topic of the opioid crisis and its impact on children		
Yes	1,132	53.0
No	1,003	47.0

93.7% of participants said that they were "likely" or "extremely likely" to continue using these skills in the future. Most respondents also indicated that they would likely use these skills in the near future, with 55.7% saying they would use the skills immediately or within a few weeks and another 23.7% indicating they would use their new knowledge within a few months.

Discussion

As hypothesized, this program evaluation found that SCOPE was effective at rapidly increasing the capacity of the interdisciplinary workforce to serve children and families affected by NAS. Specifically, SCOPE was able to be deployed rapidly

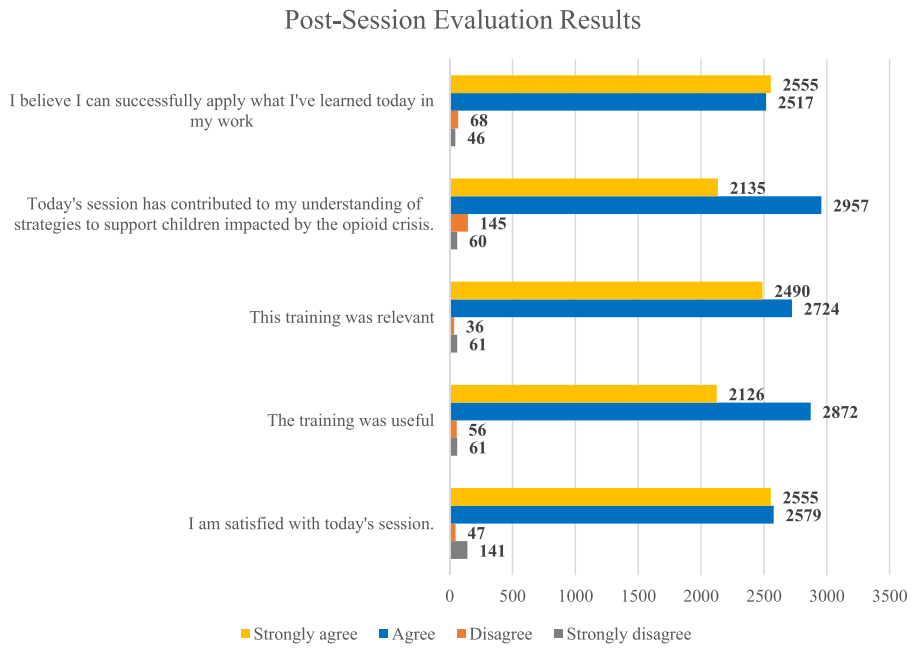

FIGURE 2
Participant self-reported understanding, satisfaction, relevance, and application after SCOPE.

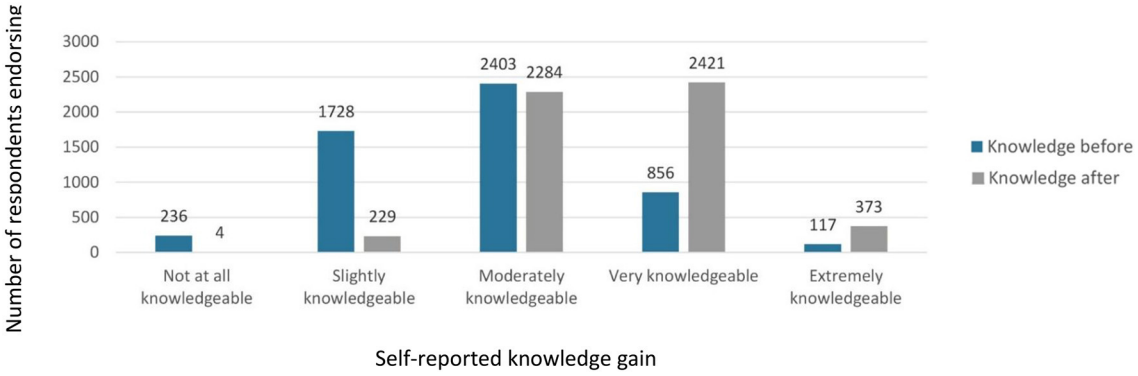

FIGURE 3
Participant self-reported knowledge before and after completion of SCOPE.

through the ECHO model and successfully increase the skills and knowledge of providers. Moreover, SCOPE allowed providers to quickly start using their newly acquired skills and has the potential to impact a large number of children at risk for developmental challenges across a broad geographic range.

One of the most striking findings of this analysis is how effectively this approach was able to quickly reach and significantly impact the NAS workforce. Specifically, participants had a large increase in knowledge over a relatively brief period of training that was at the level they said they would need to be successful. This included both their reported knowledge level as well as their direct knowledge of opioid related information. Moreover, their confidence in their ability to use their new knowledge was rated as extremely high with over half of the sample intending to use their skills within a few weeks. Further, most participants indicated that they were "likely" or "extremely likely" to continue using their new knowledge and skills. This suggests that SCOPE can quickly increase the capacity of the NAS workforce and give professionals skills that are immediately relevant to their current and future caseloads. The speed with which this program can be deployed, and its broad reach make it a valuable tool to train and support professionals managing the developmental challenges of NAS.

Given the complexity of meeting the needs of those with NAS and the multiple professions and systems involved (18), interprofessional development to expand the workforce trained to address this public health problem is critical. By specifically developing a curriculum based on emerging evidence and best practices, our NTI was able to ensure that professionals from a

range of disciplines across 14 states received the most up-to-date practice information in SUD and NAS. Overall, this model allowed novel strategies used throughout the network to be implemented broadly, thereby increasing capacity and the infrastructure needed for public health and policy initiatives.

This contrasts with many professional development strategies which are notoriously slow, costly, and do not effectively support adult learners (35). Given this, SCOPE may be a particularly valuable tool to address the major public health implications on children of the opioid crisis. This project adds to the growing body of research that demonstrates that the ECHO model is particularly effective at delivering relevant and timely information to providers that allows them to provide equally effective care as specialists [e.g., (36–38)].

Another striking feature of this project is the broad reach of SCOPE. As can be seen from the heat maps, SCOPE was able to reach an extremely diverse geographic range. This included both urban and rural areas, more compact eastern states and large western states. Participation was clearly centered around states that were part of the NTI, but participants joined from many other states as well. This suggests that professionals are searching out professional development opportunities related to NAS and that the ECHO model can support practitioners who would otherwise not have received the training offered by SCOPE. This, along with the high satisfaction scores suggest that SCOPE is a highly acceptable training model.

It is also noteworthy that participants from many counties were in rural or frontier areas. Traditionally, these areas struggle to meet clinical needs (39) and encounter persistent health care shortages (40). That SCOPE was able to train many providers from rural areas is a particular strength of this model. Indeed, ECHO was originally developed as a tool to support physicians in rural communities (41), and these data further demonstrate the success of ECHO for this purpose. In addition to rural and frontier participants, a large number of providers joined from urban areas as well. This could be due to the immense need in those communities, the ease of use of the ECHO model, or both. This demonstrates that ECHO is able to reach rural and underserved communities that traditionally struggle with access to specialty care.

The estimate of the number of children impacted by SCOPE is also striking. Although this is a self-reported estimate, and not an actual count of impacted children, this still demonstrates that SCOPE can potentially reach an extremely large number of children in need for a relatively modest investment of time. It is also worth noting that the skills professionals gained through SCOPE are applicable well into the future for other children with developmental concerns and/or complex psychosocial histories (e.g., complex traumatic stress). For example, having greater awareness of trauma (a key topic in SCOPE) may lead clinicians to be more attuned to trauma reactions in children who present for non-NAS related problems. Further, that clinicians can use these skills immediately may allow them to maintain their knowledge more effectively and make it part of their ongoing practice. Indeed, previous research suggest that to be effective, professional development needs to have opportunities to practice the skills (42). This conforms to adult learning theory (43) and is a founding principle of the ECHO model.

Results indicate that many participants work directly with children at-risk for NAS and reported increased confidence in implementing strategies, increased connection to the community of practice, high intent to utilize new knowledge and strategies on the job, and significant increases in knowledge of opioid impact on communities. Nearly all participants reported satisfaction with the sessions, and they felt that the sessions contributed to their overall understanding of strategies to support children impacted by the opioid crisis. Important to behavior change, participants reported they were utilizing new knowledge, skills, and strategies then planned to continue to use new practices in their work. Specifically, during the case discussions, evidence-based recommendations and strategies to improve outcomes of the child/family were shared so participants could implement immediately in their practice. These discussions are solution-focused and consider the resources and practices of each distinct state or region. By gaining access to a group of experts (SCOPE hub team), the participants engaged in complex conversations about the interactions of child and family characteristics, SDOH, and the systems within which they existed. They worked collaboratively, in real-time, to develop practical solutions that could then be implemented in their state/region. SCOPE, therefore, is a promising approach for increasing the capacity of providers to support children with NAS and their families, thereby increasing the capacity of our healthcare system to respond to this epidemic.

Limitations

Although we had a large sample and found compelling evidence that SCOPE is an effective program that can rapidly reach a wide range of providers, there are several limitations to note. First, due to the nature of this project, we are limited in the conclusions that can be made. Specifically, this was a program evaluation of the impact a professional training program on providers, rather than a controlled study of patient outcomes. There is no control group or alternative condition with which to compare our results. Therefore, we cannot conclude that the changes reported here are unequivocally due to SCOPE. However, given that there are limited professional development opportunities related to NAS, and none have the same focus as SCOPE, it seems possible that this program was the primary driver of this effect. Future research would be needed to make a stronger claim and to explore more nuanced reasons why some participants might not have been satisfied with the program.

Further, the implementation of SCOPE was not monitored at each site. Therefore, it is not possible to claim that there was consistency in the professional development received by all trainees. However, the ability to adapt curricula to meet local needs is a key feature of ECHO. Indeed, after the initial training of the state sites, they were all free to customize the program to suit their local needs. This ability to adapt the program could be one reason we were able to reach such a large geographic region. It may also account for some of the knowledge gains we observed. That is, providers may have been more engaged in the material because each state team was able to make the content relevant. While we

cannot confirm based on our data, this type of adaptation is a known element of successful implementation (44, 45). Regardless, it will be important to better understand the elements of this program that were critical drivers of the outcomes. Future research on SCOPE within an implementation science framework could clarify this more effectively.

Finally, given that this was a program evaluation, there were limitations in our ability to measure some outcomes directly. For instance, providers' implementation strategies were self-reported, the number of children with NAS on their workload was estimated and knowledge gains were indirectly assessed. Therefore, these findings should be interpreted with care. Nonetheless, given the large scale of this program, this is encouraging evidence that SCOPE contributed to the desired outcomes.

Conclusion

As illustrated in this project, the ECHO model is feasible, acceptable, and effective, and offers a timely approach to increase the capacity of the states and communities to meet emerging health needs, such as that of the opioid crisis. When paired with a specifically designed curriculum and a nationwide method for dissemination, it can be a powerful tool for (1) enabling equitable access, (2) building a diverse and skilled workforce, (3) improving and innovating through evaluation, research, and quality improvement, and (4) building and maintaining a strong organizational infrastructure for public health. Decision-makers and administrators are called to support and extend the use of interprofessional education and training modalities, such as ECHO, to improve family outcomes. Our findings should be used to advocate for systems-level funding to support interprofessional training and capacity building across all systems inclusive of partners across sectors.

Specific to the opioid crisis, Project SCOPE was a highly effective method of increasing the capacity of interdisciplinary providers to address cross-system issues impacting children and families. This approach provides a training modality which allowed interdisciplinary professionals to obtain continuing education on a range of developmental, social-emotional, health, and systems-level topics arming them with skills to effectively support families and children impacted by substance use disorders and related trauma. Moreover, this model can be rapidly adapted and deployed to reach a wide geographic region, especially areas that are highly affected by the opioid crisis and underserved rural communities. Further, state teams who have participated in SCOPE have the benefit of an ongoing infrastructure and local community of practice to minimize the impact of the opioid epidemic on children and families in the future. These children are increasingly noted to have physical health impacts, behavioral health concerns, and educational needs. SCOPE can effectively offer interdisciplinary professionals from cross-sector perspectives the necessary knowledge, skills, and opportunities for collaboration to prevent and/or mitigate long term sequalae of prenatal opioid exposure on children and families, while the ECHO model can serve as a promising practice for public health prevention for a range of complex health care needs or public health emergencies.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the University of Wyoming Institutional Review Board. The studies were conducted in accordance with the local legislation and institutional requirements. The Ethics Committee/Institutional Review Board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because this was a training program resulting in program evaluation. Participants self-selected into the training and were not required to complete the evaluation measures in part or full to receive the training.

Author contributions

SW: Conceptualization, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Visualization, Writing – original draft, Writing – review & editing. CH: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. AW: Conceptualization, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Visualization, Writing – original draft, Writing – review & editing. ED: Supervision, Validation, Visualization, Writing – original draft, Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software. EB: Conceptualization, Funding acquisition, Investigation, Methodology, Project administration, Resources, Visualization, Writing – review & editing. JT: Conceptualization, Data curation, Investigation, Methodology, Project administration, Resources, Visualization, Writing – original draft. TB: Data curation, Investigation, Methodology, Project administration, Resources, Supervision, Writing – original draft. AD: Data curation, Formal analysis, Software, Visualization, Writing – original draft, Writing – review & editing. EM: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

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

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Changes in LEND trainees' understanding and application of diversity, equity, inclusion, and justice principles

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Objectives: To assess changes in trainees' knowledge and application of Diversity, Equity, Inclusion, and Justice (DEIJ) concepts after participating in a midwestern academic medical center Leadership Education in Neurodevelopmental and Related Disabilities (LEND) program. LEND is a federally funded year-long program training individuals of various disciplines (e.g., speech pathology, family advocacy, psychology) to better support the health of individuals with disabilities.
Methods: Trainees ($n = 46$) answered questions about their knowledge and application of DEIJ topics before and after program participation in 2021–2022 and 2022–2023. Changes in trainees' responses were examined using paired-samples *t*-tests.
Results: Thirty-six (78%) participants identified as White, 7 (15%) as Black, 2 (4%) as Asian, and 2 (4%) as more than one race. Three (7%) participants identified as Hispanic/Latino. Over the one-year program, trainees' perceived knowledge increased [$t(45) = 5.84$, $p < .001$, $M_{diff} = .59$, *Cohen's D* = 0.86]. Regarding articulating definitions of DEIJ terms, trainees' summed scores following program participation also improved [$t(45) = 4.71$, $p < .001$, $M_{diff} = 2.37$, *Cohen's D* = 0.70]. However, their comfort with addressing prejudicial statements and discussing and combating "-isms" (application of DEIJ skills) did not increase [$t(45) = 1.74$, $p = .09$, $M_{diff} = 0.17$, Cohen's D = 0.26].
Conclusions for practice: LEND program participation positively impacted trainees' perceived DEIJ knowledge and ability to define DEIJ terms. However, future refinements to the curriculum will be needed to improve trainees' application of skills and to develop a more nuanced understanding of equity, intersectionality, inclusion, and belonging.

KEYWORDS

curriculum, leadership, developmental disabilities, inclusion, equity, intersectionality, diversity

Introduction

Large racial and ethnic disparities exist in the healthcare system, such that racially and ethnically minoritized individuals have less access to high quality healthcare (1–3), experience discrimination (4), do not receive culturally responsive care, and consequently, have poorer health outcomes (3, 5). Other minoritized populations, such as individuals with disabilities (6), individuals identifying as LGBTQIA + (7), those

without English language proficiency (8), and individuals born outside of the United States (9), also experience healthcare disparities. Though large-scale systemic changes are needed to decrease disparities, training healthcare providers to provide culturally responsive care can increase their cultural competence, improve patient satisfaction (10) and promote patients' health self-efficacy (11) among minoritized populations. Culturally responsive care is defined as care that seeks to understand and address a family's background, their belief systems, and the social contributors of health impacting the family, including institutionalized and personally mediated racism (12).

A recent review identified 89 articles examining the efficacy of training to improve cultural competency and reduce health disparities (13). Educational programs most frequently used simulations (e.g., role plays), discussion groups, lectures, immersion experiences, case-based learning, and reflections, and typically combined educational strategies. Using mixed teaching strategies was associated with greater increases in learners' knowledge and attitudes; however, most studies used self-reported assessment which may be positively biased (13, 14). In addition, few studies demonstrated improvements in participants' skills. While many studies increased learners' knowledge of racially and ethnically minoritized populations and the LGBTQIA + community, as well as encouraged reflection on their own culture and background, few of these studies specifically addressed disability.

Culturally responsive care is particularly important for individuals with disabilities, who typically interact with the healthcare system more frequently than individuals without disabilities (15, 16). Unfortunately, disability remains underrepresented in healthcare education (17, 18), leaving many healthcare providers unprepared to work with this population (19). Consequently, individuals with disabilities frequently face ableism in the healthcare system, such as a lack of physical and cognitive accessibility and discrimination (20–24). Furthermore, despite a well-documented history of unequal healthcare access for individuals with disabilities, the National Institute of Minority Health and Disparities only officially recognized people with disabilities as a population with health disparities in September 2023. The exclusion of and discrimination towards individuals with disabilities contributes to the lack of knowledge and interventions aimed at reducing disparities this population faces, particularly for individuals who also belong to other marginalized groups (e.g., racial, ethnic, sexual, and gender minorities).

Kimberlé Crenshaw introduced the concept of holding multiple marginalized identities through her coining of the term "intersectionality." Intersectionality refers to "the precise nature of discrimination that occurs when multiple axes of identity are vulnerable to oppression" (25). This concept is crucial for understanding and addressing healthcare inequities faced by individuals with disabilities, as (1) individuals with disabilities often hold identities beyond their disability (or disabilities) that shape their lived experiences, and (2) individuals with minoritized identities are more likely to have disabilities, often as a result of the discrimination and exclusion they encounter (26–28). Therefore, training aimed at promoting culturally responsive care for providers serving individuals with disabilities would benefit from incorporating intersectionality-based concepts to address the health inequities this population faces (29).

Leadership Education in Neurodevelopmental and Related Disabilities (LEND) programs are uniquely poised to train healthcare professionals in culturally responsive care for individuals with disabilities, given their focus on equity and interdisciplinary collaboration. Funded by the Maternal Child Health Bureau (MCHB), these interdisciplinary programs aim to enhance the health and well-being of individuals with disabilities across the lifespan, from infancy to adulthood. With 60 programs across the United States, LEND recruits and trains graduate-level students in fields such as medicine, psychology, and occupational therapy, as well as individuals with developmental disabilities and their family members.

LEND training is guided by the Maternal and Child Health (MCH) Leadership Competencies (30). A key competency is Diversity, Equity, Inclusion, and Accessibility, which aims to ensure that all people and communities are treated with respect, reduce barriers to equity, decrease disparities, and improve health outcomes (30). The interdisciplinary approach of LEND also ensures that the voices of individuals with disabilities and their allies are represented throughout the training process.

Studies have shown that LEND training enhances trainees' self-efficacy and leadership competencies, as observed by both faculty and trainees (31–33). Research also suggests that healthcare providers trained through LEND are more likely to work with underserved populations than those who have not participated in the program (34). However, to date, no studies have specifically examined LEND's impact on Diversity, Equity, Inclusion, and Justice (DEIJ) competencies. The current study seeks to fill this gap by evaluating how LEND curricula influence trainees' knowledge and application of DEIJ principles.

Method

This study was deemed exempt from human subjects research by Cincinnati Children's Hospital Medical Center's Institutional Review Board.

The Cincinnati Children's LEND training program

The Cincinnati Children's LEND program is a one-year program in which learners participate in over 300 h of training. Training is centered around adult learning, including didactic lectures, evidence-based case presentations, interactive/simulated learning experiences, interdisciplinary clinical care, leadership skills development, and cultural humility activities. In addition, trainees participate in eight or more hours of discipline-specific experiences per week.

The Cincinnati Children's LEND program has also continuously revised its DEIJ curriculum in response to learning from materials created by people with disabilities and other

marginalized identities and updated research about the impact of systemic inequity on people with disabilities. The DEIJ curriculum was developed using the MCH conceptual framework. The MCH Leadership Competencies are organized within a conceptual framework in a "progression from self to wider community demonstrating the widening contacts, broadening interests, and growing influence that MCH leaders can experience over their career" (30). The conceptual framework develops and progresses from "self," focusing on increasing knowledge and reflection, to "others," in which leadership extends to coworkers, colleagues, students, and patients, to the "wider community," broadening the impact of leadership to organizations and systems. The Cincinnati Children's LEND DEIJ curriculum centers around these leadership competencies, with particular emphasis on the MCH *Diversity, Equity, Inclusion and Accessibility* competency, *cultural humility* (35) and *structural competency*, which is the recognition of the economic, political, and social factors that produce health disparities and contribute to disease (36).

In line with the MCH Leadership Competencies and conceptual framework, the following DEIJ-related training goals were developed: "At the end of the training year, trainees should be able to (1) define equity in healthcare, (2) describe historical roots that drive inequity today as well as current policies and practices that maintain inequity, (3) identify the impact of their intersecting identities and privilege on lived experiences and interactions with others, (4) examine their own implicit bias, and (5) identify ways to address systemic inequality in healthcare."

Throughout the year, trainees participated in five, 2-h DEIJ sessions. Each DEIJ session was led by professionals with extensive knowledge and teaching experience in the field of DEIJ, many from marginalized populations themselves and/or who were also trained in the LEND program. Some session leaders are LEND faculty while others are outside consultants. In the first session, which focuses on knowledge building and self-reflection, trainees reflected on their intersecting identities, considering how their identities afford or do not afford them privilege. To build a foundational knowledge base, the second session focused on developing greater awareness of inequity in healthcare and ways to increase equitable care (e.g., person-centered care). The third session continued to emphasize self-reflection, while broadening the sphere to "others," describing implicit and explicit bias and providing trainees with microintervention strategies to respond to microaggressions. The fourth and fifth sessions addressed the "wider community" by asking students to reflect on community, systems-level, and policy changes needed to increase access to and quality of health care. DEIJ content was also infused into the curriculum throughout the year (see Table 1 for examples).

We used evidence-based teaching methods to promote growth in the four major tenets of cultural humility (see Table 2). Aligned with previous successful curricula, we paired lectures with active learning strategies, such as group discussions and reflection activities (13). For example, in a lecture discussing how to create accessible presentations, trainees were presented with case scenarios of groups for which they would design an accessible

TABLE 1 Examples of DEIJ throughout LEND curriculum.

Training domain	Lecture title	Objectives	Activities
Leadership: Individual and team-based leadership skill development	Values in Disabilities	Recognize history of disability rights and ways attitudes towards disability have changed over time	Small and large group reflection—students reflected on disability representation in the media
Core curriculum: Broad overview of disability topics	Intellectual and developmental disabilities	Describe different conceptualizations of intellectual and developmental disabilities	Small and large group reflection—Students reflected on a TEDTalk by a Black woman with an intellectual disability, who described her experiences of marginalization and resilience
Seminar in Evidence Based Methods: Team-based research projects	Quantitative methods	Identify the research cycle, ways biases impact research, and the importance of partnering with communities to engage in research	Article review—Students critically reviewed a peer-reviewed journal article to identify the impact of racism on research questions, methods, data analysis, and interpretation
Evidence-Based Case Discussions: Series of seven case series scenarios	ADHD, Down Syndrome, Fragile X, Developmental Coordination Disorder, Cortical Visual Impairment, Spina Bifida	Understand the prevalence and impact of different diagnoses on individuals and families; reflect on the lived experiences of individuals with different conditions	Journaling exercise—Students critically examined peer-reviewed journal article for each lecture and provided written responses describing the impact of neighborhood, community factors/resources, and social factors (e.g., discrimination) on individuals with this type of condition or disability

TABLE 2 Four tenets of cultural humility addressed in the LEND curriculum.

Tenet	Definition
Awareness[a]	Awareness or insight into trainees' privileged identities, marginalized identities, biases, and communication with culturally diverse individuals
Attitudes	Understanding personal values and belief systems and the way in which upbringing and lived experiences shape them
Knowledge	Awareness of the impact of systemic inequity on marginalized families and knowledge of one's own behaviors when interacting with those from a different culture
Skills	Integrating cultural humility and advocacy for equity in daily practice (e.g., asking families about structural barriers)

[a]Definitions adapted from Tervalon, Murray-Garcia to reflect learning objectives of LEND.

presentation (e.g., a group of deaf students, a group of racially diverse family advocates). Students first reflected on ways to adapt the presentation and subsequently discussed ideas for adaptation in small groups.

In addition, we used case-based learning, including de-identified case vignettes and personal case presentations from families of children with disabilities to increase trainees' knowledge and awareness (37–39). Videos were used to amplify the voices of marginalized individuals, with videos demonstrating individuals' perspectives and lived experiences. We also played videos of experts discussing research and providing information on topics such as equity, environmental racism, disability justice, and the school-to-prison pipeline. Videos, in combination with other educational strategies, have been associated with greater knowledge (40–43), awareness (40, 43, 44), and skills (13). Trainees also periodically prepared for sessions with readings or read short articles during sessions, including research articles related to case-based discussions and articles sharing individuals' experiences (45). Throughout the year, sessions incorporated educational technology, such as polls and live quizzes, to increase reflection and engagement (46).

Measures

Trainees were asked to complete the following measures before and after participation in the LEND program. All survey responses were collected through REDCap and were anonymous. To seek iterative feedback for program improvement, pre-post assessment surveys are a standard component of LEND. Therefore, participants were not compensated for completing the measures for this study. Trainees first completed a demographic survey to obtain information about their race, ethnicity, and other sample characteristics. Due to the lack of validated measures in this area, the following measures were created for this study.

DEIJ perceived knowledge and application

We asked participants to indicate to what extent they agreed with nine statements related to understanding and combating structural inequity (e.g., racism, sexism), on a 6-point Likert scale from 1="strongly agree" to 6="strongly disagree." Similar to the DEIJ terms, authors created these questions because they are direct targets of the LEND DEIJ training goals and are addressed in the curriculum. The questions were developed by LEND faculty and staff (AF, SW, JS) and reviewed by the department's anti-racism training program, who are trained in DEIJ education and implement DEIJ training curriculum across Cincinnati Children's Division of Developmental and Behavioral Pediatrics (TF, NR, SA). Six of the nine statements asked trainees about their perceived understanding of biases, equity, culture, privilege, intersectionality, and ableism and were considered Perceived Knowledge prompts. Three of the nine statements assessed trainees' perceived comfort with confronting and discussing prejudice and discrimination and were considered Application prompts. Separate mean Perceived Knowledge and Application domain scores were created by summing the scores for each item in a domain and then dividing by that domain's number of items. A mean Perceived Knowledge and Application total score was created by summing the scores for all nine prompts and dividing by nine.

DEIJ knowledge

Trainees were asked to define nine DEIJ terms: equity, privilege, intersectionality, ableism, microaggressions, systems-centered language, diversity, inclusion, and belonging. They were given the choice to select the following response options, "I am unsure what this word means," and "I have not heard of this word." The authors chose these terms because of their importance and relevance to culturally responsive leadership and because they are targets of the LEND DEIJ training goals. Therefore, these concepts were addressed throughout the LEND curriculum. For example, we discussed "belonging," creating a safe space where individuals with disabilities can be their authentic selves, in several LEND training sessions.

The Knowledge of DEIJ terms measure was scored by comparing trainee definitions of DEIJ terms to standard definitions for each item (47). Responses were marked as "correct" (2 points), "partially correct" (1 point), and "incorrect" (0 points), similar to scoring on the Weschler Intelligence Scale for subtests assessing individuals' vocabulary and verbal reasoning (48). Responses were scored as "partially correct" if they included one or more aspects of the correct definition but did not provide a complete and thorough definition. For example, if a trainee stated that "privilege" was an advantage but did not include that privilege is not earned, the definition was marked as "partially correct."

Participants

Trainees answered questions before and after completion of the LEND program in the 2021–2022 ($n = 24$) and 2022–2023 ($n = 22$) training year.

Data analysis

Because there were no differences between trainees' responses in 2021–2022 and 2022–2023 on the outcome measures, we combined responses across the two LEND training years. We used paired-samples t-tests to examine pre-post changes in the Perceived Knowledge and Application total score (average of all nine questions) and average "Perceived Knowledge" and "Application" domain scores.

For the knowledge of DEIJ terms, two independent coders from the research team evaluated and scored trainees' responses to each term. They met and came to consensus on discrepant ratings without disagreement. A third coder reviewed 25% of trainees' responses for inter-rater reliability in year 1 and 10% in year 2, which was substantial (Cohen's Kappa = .73). Trainees' scores on each of the nine terms were summed to create a Knowledge total score.

To assess changes in Knowledge of DEIJ terms, we summed trainees' response scores for each term at the pre- and post survey administration and conducted a paired-samples *t*-test to examine changes in trainees' total scores across the nine terms. We described trainees' "post-LEND" definition responses to better identify trainees' understanding after participating in the LEND training program and where changes to the curriculum may be warranted to improve trainees' knowledge of DEIJ terms.

Results

Most participants were White (78.3%), female (91.3%), and came from a variety of disciplines (see Table 3). Five (11%) trainees identified as having a disability, one (2.2%) as having a special healthcare need, eight (17.5%) as a parent of a child with a disability or special healthcare need and 15 (33%) as a non-parent family member of a person with a disability or special health care need (e.g., sibling of a person with a disability).

Mean scores on the Perceived Knowledge and Application of DEIJ (Table 4) increased across the LEND training year [$t(45) = 4.94$, $p < .001$, $M_{diff} = .45$, *Cohen's* $D = 0.73$]. While trainees' Perceived Knowledge increased [$t(45) = 5.84$, $p < .001$, $M_{diff} = .59$, *Cohen's* $D = 0.86$], trainees' comfort with addressing prejudicial statements and combating "-isms" (Application) did not increase over the LEND year [$t(45) = 1.74$, $p = .09$, $M_{diff} = 0.17$, Cohen's D = 0.26].

Trainees' summed scores following participation in LEND increased [$t(45) = 4.71$, $p < .001$, $M_{diff} = 2.37$, *Cohen's* $D = 0.70$; See Figure 1]. Before LEND, 37% of trainees' definitions were given 0 points in comparison to 19% after LEND. The percentage of trainees' responses labeled "partially correct" increased from 34% before LEND to 45% after LEND. The percentage of trainees' responses labeled "correct" increased from 28% to 36%.

When examining responses to individual terms, most trainees ($n = 26$, 57%) described what "equity" is (i.e., fair distribution of resources based on individual need) after LEND participation; however, only seven (15%) trainees gave a 2-point response, acknowledging why equity is necessary (i.e., equity is needed to prevent structural inequality leading to oppression). Similarly, half of trainees described "privilege" as an advantage ($n = 23$, 50%), whereas 20 (44%) trainees gave a 2-point response,

TABLE 3 Demographic characteristics of the sample.

Characteristic, N (%)	2022, *n* = 24	2023, *n* = 22	Total sample
Gender			
Female	22 (92%)	20 (91%)	42 (91.3%)
Male	1 (4%)	2 (9%)	3 (6.5%)
Non-binary	1 (4%)	0 (0%)	1 (2.2%)
Race			
White	19 (79%)	17 (74%)	36 (78.3%)
Black/African American	3 (8%)	4 (17%)	7 (15.2%)
Asian	1 (4%)	1 (4%)	2 (4.3%)
More than one race	1 (4%)	1 (4%)	2 (4.3%)
Ethnicity			
Hispanic/Latino	2 (8%)	1 (4%)	3 (6.5%)
Non-Hispanic	22 (92%)	21 (96%)	43 (93.5%)
Education			
Certification program	1 (4%)	0 (0%)	1 (2.2%)
Associate's degree	0 (0%)	2 (9%)	2 (4.3%)
Bachelor's degree	9 (36%)	8 (36%)	17 (39.5%)
Master's degree	10 (40%)	9 (40%)	19 (41.3%)
Medical/Doctoral degree	4 (28%)	3 (13%)	7 (15.2%)
Discipline			
Psychology	4 (16%)	5 (21%)	9 (19.6%)
Developmental-Behavioral Pediatrics	3 (12%)	2 (9%)	5 (10.9%)
Speech-Language Pathology	2 (8%)	2 (9%)	4 (8.7%)
Genetic Counseling	2 (8%)	2 (9%)	4 (8.7%)
Family advocacy	2 (8%)	3 (13%)	5 (10.9%)
Adult Services	1 (4%)	0 (0%)	1 (2.2%)
Advocacy	1 (4%)	1 (4%)	2 (4.3%)
Applied Behavior Analysis	1 (4%)	0 (0%)	1 (2.2%)
Audiology	1 (4%)	1 (4%)	2 (4.3%)
Disability Studies	1 (4%)	0 (0%)	1 (2.2%)
Education	1 (4%)	0 (0%)	1 (2.2%)
Family Medicine	1 (4%)	0 (0%)	1 (2.2%)
Occupational Therapy	1 (4%)	2 (9%)	1 (2.2%)
Physical therapy	1 (4%)	1 (9%)	2 (4.3%)
Public Health	1 (4%)	1 (4%)	2 (4.3%)
Social Work	1 (4%)	2 (9%)	3 (6.5%)

TABLE 4 Pre- and post-scores on the perceived knowledge and application of DEIJ survey.

Perceived knowledge domain prompt	Pre, mean (SD)	Post, mean (SD)
"I am aware of biases that I have towards individuals with backgrounds different from my own"	4.91 (1.02)	5.33 (.60)
"I can identify -isms (such as racism, classism, Eurocentrism) that are used in policies and by institutions (e.g., government)."	4.63 (.95)	5.30 (.70)
"I am aware of my identities that give me privilege."	5.07 (.98)	5.30 (.89)
"I understand the ways in which my cultural background influences my views of the world."	5.07 (.84)	5.43 (.54)
"I understand intersectionality within the disability community."	4.46 (1.57)	5.24 (.67)
"I understand the impacts of ableism on individuals with disabilities' everyday lives."	4.43 (1.39)	5.46 (.59)
Perceived Knowledge Domain Average	4.76 (.71)**	5.35 (.45)**
Application Domain Prompt	**Pre, Mean (SD)**	**Post, Mean (SD)**
"I feel comfortable confronting prejudicial statements and microaggressions in my everyday life."	4.52 (1.13)	4.89 (.90)
"I am committed to fighting against -isms (such as ethnocentrism, heterosexism, genderism) in my everyday life."	5.59 (.65)	5.54 (.62)
"I feel comfortable discussing -isms (such as ageism, sexism, ableism) in my everyday life (e.g., personal, professional, with family, with friends).	5.02 (1.11)	5.24 (.77)
Application Domain Average	5.05 (.77)	5.22 (.62)
Perceived Knowledge and Application Average	4.86 (.64)**	5.31 (.45)**

**$p < .001$.

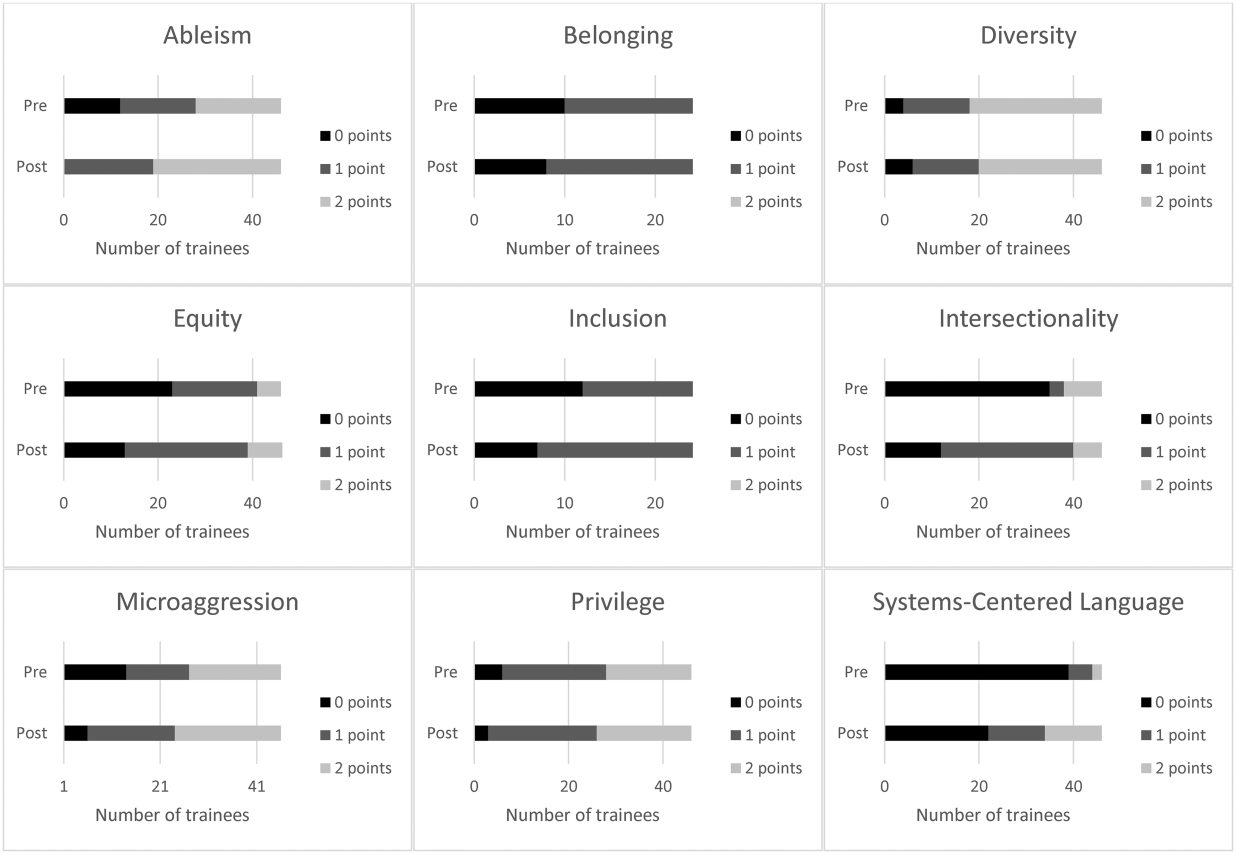

FIGURE 1
Number of 0-point, 1-point, and 2-point responses to trainees' Diversity Equity Inclusion and Justice term definitions before and after Leadership Education in Neurodevelopmental and Related Disabilities.

describing that privilege is an advantage that is unearned or innate. With "intersectionality," 28 (61%) trainees reported that intersectionality is an interaction or intersection of identity groups; however, only six (13%) trainees acknowledged that the intersection of identities leads to privilege and/or marginalization (2-point response). Most trainees ($n = 27$, 59%) provided a 2-point response for "ableism" though the remaining 19 (41%) trainees did not provide thorough descriptions of ableism. For example, one trainee described that ableism is discrimination against individuals with intellectual disability, thereby incorrectly excluding other types of disabilities in their response. Twenty-two (48%) trainees provided 2-point responses when asked to define "microaggressions." The remaining 24 trainees (52%) provided incorrect definitions (e.g., "passive aggressive") or partially correct responses (e.g., did not acknowledge that microaggressions cause harm, did not differentiate microaggression from macroaggression). Many trainees ($n = 22$, 48%) were unable to define "systems-centered language" after LEND. Twelve (26%) understood that systems-centered language involves language that describes systems rather than individuals, but only 12 (26%) trainees described that the term holds systems accountable for perpetrating oppression rather than blaming individuals for existing disparities (2-point response). With "diversity," 14 (30%) trainees did not provide thorough definitions of the term, such as referring to but not defining "differences" (e.g., answer limited to "people being different from one another and celebrating our differences"). Nearly half of the trainees described "inclusion" as including people of different backgrounds and/or ability statuses ($n = 22$, 48%); seventeen (37%) trainees described that it is important to ensure the environment or space allows for participation or inclusion (2-point response). Finally, regarding "belonging," 26 (57%) trainees expressed the inward feeling of being a part of a group or being able to be one's authentic self. Similar to "inclusion," only 12 (26%) trainees gave a 2-point response, describing the importance of the environment or community's contribution to that sense of safety.

Discussion

We found that trainees' perceived knowledge about DEIJ increased after participation in LEND, with a large effect size. Before LEND participation, trainees were likely to "Somewhat Agree" they understood important DEIJ concepts, and after LEND, trainees were likely to "Agree" they understood important

DEIJ concepts. Similar to previous research, we did not identify an increase in trainees' comfort with application of DEIJ [i.e., addressing systemic inequity and prejudice in their everyday lives (13)]. Trainees responded on average between a 5 ("Agree") and 6 ("Completely Agree") on a 6-point Likert scale to two of the three application-based questions, possibly reflecting social desirability bias, which is common in scales assessing cultural humility (14). The high baseline scores may contribute to a ceiling effect, limiting the potential for measurable improvement. Additionally, the application questions asked trainees about their comfort in addressing prejudicial statements and "-isms" in their everyday life, which can be influenced by many factors, including power differentials, fear of negative consequences and retaliation, and being in an unsafe culture or environment. It may be helpful to explore when and how trainees confront microaggressions, prejudicial statements, and inequity through qualitative interviews or observation. Though the LEND program uses case-based learning, small group discussions, and role play to promote behavior change, trainees need additional practice and experience to increase their comfort with applying DEIJ concepts in their everyday life.

When defining DEIJ terms, trainees' overall knowledge increased, with a medium effect size. Most trainees gained a basic understanding of important DEIJ terms, but only about one-third of responses were in-depth, complete definitions following LEND program participation. Trainees appeared to be most knowledgeable about ableism, diversity, and microaggressions after LEND completion, with 50% or more trainees providing 2-point definitions for these responses. By contrast, less than one-third of trainees provided 2-point responses for equity, intersectionality, systems-centered language, inclusion, and belonging. For these terms, trainees often failed to articulate systemic implications. For example, many trainees did not describe that equity is needed to mitigate the impacts of systemic oppression, that the intersection of identity leads to privilege or marginalization, or that the environment and culture need to shift to facilitate inclusion and belonging. These findings highlight that the LEND program needs to focus more on structural competency to facilitate trainees' understanding of systemic factors that contribute to oppression.

This study should be considered in the context of its limitations. First, the cohort's demographic characteristics (i.e., the LEND trainees) may have influenced our findings. Most trainees were White women with some graduate training, pursuing service careers dedicated to working with individuals with disabilities. Therefore, most trainees have multiple privileged identities, and their career interests may increase their motivation to learn about DEIJ, especially as it relates to better supporting diverse individuals with disabilities.

Second, there are few validated assessment measures to understand individuals' knowledge and application of DEIJ. As such, we developed measures for use in this program. Future research should assess the psychometric properties of our measures, with a larger sample size. We addressed some limitations of previous self-report based studies by rating changes in trainees' understanding of important DEIJ concepts. However, we also used self-reported measures of changes in knowledge and skills. It is possible trainees responded more positively about their knowledge, especially during the post-assessment, because they were aware that the training program was designed to enhance these competencies over the year (49). Additionally, though trainees' understanding of DEIJ terms increased throughout the training year, trainees continued to demonstrate gaps in their knowledge of how structural inequality is related to systems-centered language, intersectionality, equity, inclusion, and belonging. Trainees' gaps in knowledge will be used to continually modify the LEND training curriculum to improve trainees' understanding of DEIJ.

A major limitation of the study we hope to address in future studies is identifying whether changes in knowledge of DEIJ concepts, self-reported knowledge, and self-reported skills translate to changes in behaviors, leadership or patient care. As we adapt the curriculum, we can assess increases in trainees' ability to provide culturally responsive care and skills in addressing microaggressions and structural inequality using observational methods (e.g., observations of patient encounters, team meetings) and patient-reported measures. Finally, our study examined only short-term gains in knowledge and application, and we plan to assess if improvements are maintained over time.

Conclusion

The LEND curriculum focuses on developing MCH competencies, which are designed to progress from self-awareness and reflection to leadership skills that can be applied to trainees' communities and at the systems-level. Our study demonstrated that the LEND training program increased trainees' perceived knowledge of DEIJ and ability to define important DEIJ terms and concepts. We aim to continuously improve the LEND curriculum to support trainees' skills in addressing structural inequality. LEND trainees are tomorrow's leaders who will be responsible for the design and implementation of interventions, programs, and research that address health equity. Although achieving health equity requires systems level change, these changes cannot be accomplished without starting at the individual, trainee level. Individuals with disabilities and their families are marginalized and face discrimination in healthcare. Essential to driving progress in this area is effective measurement of workforce development and training outcomes. Our study is the beginning of systematic efforts to improve trainees' cultural responsiveness. Future iterations of this study will examine the validity of our measures, use objective outcomes to examine changes in trainees' behaviors, and collaborate with sites across the LEND Network.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements.

Author contributions

AF: Writing – review & editing, Writing – original draft, Validation, Supervision, Resources, Project administration, Methodology, Investigation, Funding acquisition, Formal Analysis, Data curation, Conceptualization. LG: Writing – review & editing, Writing – original draft, Formal Analysis. SW: Writing – review & editing, Writing – original draft, Project administration, Investigation, Funding acquisition, Formal Analysis, Data curation, Conceptualization. TF: Writing – review & editing, Writing – original draft, Validation, Supervision, Resources, Project administration, Methodology, Conceptualization. SA: Writing – review & editing, Writing – original draft, Methodology, Formal Analysis, Data curation, Conceptualization. NR: Writing – review & editing, Writing – original draft, Supervision, Methodology, Formal Analysis, Data curation. JS: Writing – review & editing, Writing – original draft, Supervision, Resources, Project administration, Methodology, Investigation, Funding acquisition, Formal Analysis, Data curation.

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Conflict of interest

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

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Analyzing special needs reports for children: sociodemographic trends, diagnoses, and support areas over five years (2019–2024)

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Objective: This study aims to evaluate the special needs reports for children (SNRC) in terms of sociodemographic characteristics, diagnoses, and areas of special needs in the five years from 2019, when special health boards for children were established with the new diagnostic and evaluation criteria.
Methods: This descriptive study was conducted by retrospectively reviewing the board evaluation reports of children aged 0–18 years who applied to Batman Training and Research Hospital SNRC Health Board between March 2019 and March 2024. The study included 420 children for whom all the data in the evaluation reports could be accessed. All data of the participants were collected and analyzed through the Hospital Information Management System (HIMS).
Results: The mean age at the time of health board admission was 7.41 years with 58.3% boys and 41.7% girls. When the level of special needs was analyzed, the highest rate of 47.1% was found to have Special Condition Requirement Needs (SCRN). The most common reason for application in both boys (68.6%) and girls (64%) was to benefit from Disability Rights. In terms of the distribution of disease areas, the highest rate was in the Cognitive Development Area with 36.2%, the second highest rate was in the Movement Development Area with 27.1%, and the lowest rates were in the Genitourinary System - Surgery Area with 0.2% and in the Hematology-Oncology Area with 0.2%. When the areas of Specific Learning Disabilities (SLD) were analyzed, it was determined that the highest rate was in the area of rehabilitation/early support (intervention) requirement to support cognitive functions with 43.1%. When the special needs levels were analyzed according to the disease areas, the highest rate was found in 13 areas except for the Nephrology area and Genitourinary System-Surgery Area was SCRN. In Nephrology, the rates were equally shared between SCRN and Significant special needs (SSPD) at 50%, while the highest rate in the Genitourinary System-Surgery Field was Special needs (SEN) with 60.6%.

Conclusion: Our research emphasizes the crucial role of SNRC health board reports in tackling the pressing challenges faced by children and their families. Through harnessing these reports, we can make significant progress in identifying and supporting children with special needs, ultimately enhancing their quality of life. Our findings emphasize the influence of gender and the typical age of 7–8 years for initial evaluation. Looking ahead, it is vital to develop comprehensive strategies to raise community awareness and equip healthcare professionals with the necessary tools to optimize support systems. Through ongoing analysis and adaptation, these efforts hold the promise of fostering a more inclusive and supportive environment for children with diverse needs.

KEYWORDS

SNRC, special needs, health board, child, adolescent

1 Introduction

Special need is a state of biopsychosocial inadequacy manifested by impairment and limitation in physical, mental, psychological, and social abilities due to various reasons that occur congenitally or acquired (1). Individuals with special needs have difficulty in performing and maintaining ordinary daily activities without assistance and in adapting to social life at an acceptable level (2). Children with special needs are children who have a higher number and intensity of needs compared to their peers due to their physical, physiological, mental, or behavioral differences. The inadequacies of these children in their adaptation skills and in meeting daily needs cause them to need regular counseling, protection, care, or rehabilitation services (3).

According to World Health Organization data, 15.3% of the world population in all age groups and 5.2% of children under the age of 14 are disabled. According to the Turkey Disability Survey, 12.29% of the population in Turkey is "disabled". Under 19 years of age, the rate of orthopedic, visual, hearing, speech, and mental disability was 3.5%, while the rate of disability was 8.8% when other internal and psychiatric diseases were included (1, 4).

While adults and children were previously evaluated by the same health board in Turkey, special health boards for children were established in hospitals with the regulation that entered into force on February 20, 2019. In line with this regulation, which aims to determine the special needs status of children instead of the disability rate, the Special Needs Report for Children (SNRC) is evaluated and decided by these newly established boards (5).

For children with special needs to benefit from some economic rights such as health, care, special education, and tax reduction, it is necessary to apply to SNRC health boards with the guidance of physicians, teachers, rehabilitation centers, or with the direct request of families (4). The gains to be obtained with the result of the health committee can enable children with special needs and their families to be more involved in normal life. In this way, the birthright of every individual to be in social life can be fully maintained for children with special needs (6). In addition, today, laws, practices, and regulations for individuals with special needs are considered as one of the development indicators of countries (2).

This study aims to examine SNRC reports in the five years from 2019, when special health boards for children were established with the new diagnostic and evaluation criteria, and to draw conclusions that can guide both legislators and health professionals regarding special needs. Thanks to this awareness that will be spread throughout society, it may be possible for individuals with special needs to maintain their lives most easily and adapt to the social and cultural environment.

2 Materials and methods

This descriptive study was conducted by retrospectively screening the board evaluation reports of children aged 0–18 years who applied to the Batman Training and Research Hospital SNRC Health Board between March 2019 and March 2024. Approval for the study was obtained from Batman Training and Research Hospital Scientific Research Ethics Committee with the decision dated January 25, 2024, and numbered 375.

All data of the participants were collected and analyzed through the Hospital Information Management System (HIMS).

SNRC Health Board has 6 permanent members, including the chairman, who is a specialist in Pediatrics, and the authorized physician of SNRC. The branches of the board are Pediatrics, Child and Adolescent Mental Health and Diseases, Ophthalmology, Physical Medicine and Rehabilitation, and Ear, Nose and Throat Diseases.

Levels of special needs:

- No special requirements
- Special needs (SEN): 20–39
- Mild special needs: 40–49
- Moderate special needs: 50–59
- Advanced special needs: 60–69
- Very severe special needs: 70%–79% (considered severely disabled)
- Significant special needs (SSPD): 80%–89% (considered to be severely disabled)
- Special Condition Requirement (SCRN): 90%–99% (considered to be severely disabled)

Reasons for Application:

- Benefiting from disability rights
- To benefit from home care services
- Benefiting from special education services
- For children, a medical board report stating the situation due to terrorism, accident, and injury
- For caregiver salary (benefiting from Law No. 2828)

Disease Areas:

- Cognitive Development Area
- Child and Adolescent Psychiatry
- Speech-Language-Communication Development
- Endocrine System Area
- Genitourinary System - Surgical Area
- Visual Function Area
- Movement Development Area
- Hematology-Oncology Area
- Hearing Function - Ear, Nose, and Throat Area
- Hereditary-Natal Diseases Area
- Heart, Circulatory System Area
- Metabolism Area
- Nephrology Area
- Digestive System Area
- Nervous System Area
- Burns Area

In addition to the child's disability, the Board decided on the level of special needs and the reasons for application. Diagnoses were determined according to the ICD-10 code. Psychiatric diagnoses were made by child and adolescent psychiatrists according to DSM-5. In children older than 6 years of age, the Wechsler Intelligence Scale for Children (Wechsler Intelligence Scale for Children-R) was administered by experienced psychologists to determine the level of intelligence, and the Specific Learning Disability (SLD) battery was administered in cases where Specific Learning Disorder (SLD) was considered. In children younger than six years of age, Denver II or Ankara Developmental Screening Inventory (ADSI) administered by child development specialists was used (1).

SLD is categorized as follows:

- SLD1: Need for rehabilitation/early support (intervention) to support cognitive functions
- SLD2: Need for physiotherapy, occupational therapy, rehabilitation
- SLD3: Need for devices, orthoses, prostheses, wheelchairs, and other equipment
- SLD4: Speech and language therapy/rehabilitation requirement
- SLD5: Need for therapy/rehabilitation for hearing impairment/loss
- SLD6: Therapy/rehabilitation requirement for visual function limitation/loss
- SLD7: Therapy/rehabilitation needs for autism spectrum disorder
- SLD8: Therapy/rehabilitation requirement for specific learning disabilities
- SLD9: Need for rehabilitation at home or in hospital
- SLD10: Other

Inclusion Criteria:

- Age between 0 and 18 years
- Completion of application to SNRC
- Accessibility of complete file information

Exclusion Criteria:

- Patients with incomplete information in their files are excluded.

2.1 Statistical analysis

The data in the study were analyzed using the SPSS program (Statistical Package for Social Sciences) 25.0. Continuous data were expressed as mean ± standard deviation, median, minimum, and maximum. Pearson's Chi-Square test was used to compare categorical data. Since the study was descriptive, no further comparisons were made. Statistical significance was accepted as $p < 0.05$.

3 Results

The reports of 453 children who applied to the SNRC Health Board, which was established specifically for children within five years, were analyzed. A total of 420 (92.7%) children aged 0–18 years with complete file information were included in the study. The 33 (7.3%) children for whom all the data in the evaluation reports could not be accessed were excluded from the study. Of the 420 patients included in the study, 58.3% were boys and 41.7% were girls. The mean age of the patients was 7.41 years (±4.84 SD), the median age was 7 years, the minimum age was 0 and the maximum age was 17 years. When the special needs levels of the children were analyzed, it was seen that the highest rate was 47.1% in the "Special Condition Requirement (SCRN): 90%–99% (considered to be severely disabled)" group, followed by 26.7% in the "Special needs (SCRN): 20%–39%" and 8.3% in the "Moderate special needs: 50%–59%" groups (Table 1).

When the special needs levels of the children included in the study were analyzed separately according to gender, it was seen that the highest rate of 44.9% in boys was "Special Condition Requirement (SCR): 90%–99% (considered to be severely disabled)", followed by "Special needs (SCR): 20%–39%" with 31.4% and "Moderate special needs: 50%–59%" with 6.9%. When the distribution of girls was analyzed, it was seen that the highest rate was "Special Condition Requirement (SCRN): 90%–99% (considered to be severely disabled)" with 50.3%, followed by "Special needs (SCRN): 20%–39%" with 20% and "Moderate special needs: 50%–59%" with 10.3% (Table 2).

When the reasons for the application of the children included in the study were analyzed separately by gender, it was seen that the highest rate of both boys (68.6%) and girls (64%) was "To Benefit from Disability Rights" (Table 3).

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TABLE 1 General distribution.

		n	%
Gender	Male	245	58.3
	Girl	175	41.7
	Total	420	100,0
Special requirement level	No special requirements	25	6.0
	Special needs (SEN): 20–39	112	26.7
	Mild special needs: 40–49	10	2.4
	Moderate special needs: 50–59	35	8.3
	Advanced special needs: 60–69	6	1.4
	Very severe special needs: 70%–79% (considered severely disabled)	17	4.0
	Significant special needs (SSPD): 80%–89% (considered to be severely disabled)	17	4.0
	Special Condition Requirement (SCRN): 90%–99% (considered to be severely disabled)	198	47.1
	Total	420	100.0
Application causes	Benefiting from disability rights	280	66.7
	To benefit from home care services	112	26.7
	Benefiting from special education services	26	6.2
	For children, a medical board report stating the situation due to terrorism, accident, and injury	1	0.2
	For caregiver salary (benefiting from Law No. 2828)	1	0.2
	Total	420	100.0
Age	Mean - SD	7.41 (4.84)	
	Min-Maks-Median	0–17–7	

When the distribution of disease areas was analyzed, it was determined that the highest rate was observed in the Cognitive Development Area (36.2%), while the lowest rates were observed in the Genitourinary System - Surgery Area (0.2%) and Hematology-Oncology Area (0.2%) (Table 4).

The distribution of the patients participating in the study in the areas of SLD is shown in the table. According to this; it was observed that the highest rate was in the field of SLD1 (43.1%). It was determined that there was no patient in the area of SLD10 (Table 5).

Pearson's Chi-Square test was performed to determine whether there was a statistical difference between the gender of the participants and their SLD status. According to the test results; no statistically significant relationship was found between gender variable and SLD1 ($p = 0.188$), SLD2 ($p = 0.082$), SLD4 ($p = 0.959$), SLD5 ($p = 0.139$), SLD6 ($p = 0.875$), SLD8 ($p = 0.404$) and SLD9 ($p = 0.153$). On the other hand, a statistically significant relationship was found between the gender variable and SLD3 ($p = 0.002$) and SLD7 ($p = 0.013$). While the "None" group was higher in boys than the expected value and the "Present" group was higher in girls than the expected value for the SLD3 variable, it was concluded that the "None" group was higher in girls than the expected value and the "Present" group was higher in boys than the expected value for the SLD7 variable (Table 6).

TABLE 2 Distribution of special needs levels.

	Male		Girl		X^2	*p*
	n	%	*n*	%		
No special Total requirements	14	% 5.7	11	% 6.3	8.130	0.321
Special needs (SEN): 20–39	77	% 31.4	35	% 20.0		
Mild special needs: 40–49	6	% 2.4	4	% 2.3		
Moderate special needs: 50–59	17	% 6.9	18	% 10.3		
Advanced special needs: 60–69	3	% 1.2	3	% 1.7		
Very severe special needs: 70%–79% (considered severely disabled)	8	% 3.3	9	% 5.1		
Significant special needs (SSPD): 80%–89% (considered to be severely disabled)	10	% 4.1	7	% 4.0		
Special Condition Requirement (SCRN): 90%–99% (considered to be severely disabled)	110	% 44.9	88	% 50.3		
Total	245	% 100.0	175	% 100.0		

TABLE 3 Distribution of reasons for application.

	Male		Girl		X^2	*p*
	n	%	*n*	%		
Benefiting from disability rights	168	% 68.6	112	% 64.0	5.680	0.224
To benefit from home care services	58	% 23.7	54	% 30.9		
Benefiting from special education services	18	% 7.3	8	% 4.6		
For children, a medical board report stating the situation due to terrorism, accident, and injury	1	% 0.4	0	% 0		
For caregiver salary (benefiting from Law No. 2828)	0	% 0	1	% 0.6		
Total	245	% 100.0	175	% 100.0		

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TABLE 4 Distribution of disease areas.

		n	%
Cognitive development area	No	268	63.8
	There is	152	36.2
	Total	420	100.0
Child and adolescent psychiatry	No	358	85.2
	There is	62	14.8
	Total	420	100.0
Speech-language-communication development	No	387	92.1
	There is	33	7.9
	Total	420	100.0
Endocrine system area	No	390	92.9
	There is	30	7.1
	Total	420	100.0
Genitourinary system - surgical area	No	419	99.8
	There is	1	0.2
	Total	420	100.0
Visual function area	No	392	93.3
	There is	28	6.7
	Total	420	100.0
Movement development area	No	306	72.9
	There is	114	27.1
	Total	420	100.0
Hematology-oncology area	No	419	99.8
	There is	1	0.2
	Total	420	100.0
Hearing function - ear, nose, and throat area	No	395	94.0
	There is	25	6.0
	Total	420	100.0
Hereditary-natal diseases area	No	360	85.7
	There is	60	14.3
	Total	420	100.0
Heart, circulatory system area	No	416	99.0
	There is	4	1.0
	Total	420	100.0
Metabolism area	No	418	99.5
	There is	2	0.5
	Total	420	100.0
Nephrology area	No	416	99.0
	There is	4	1.0
	Total	420	100.0
Digestive system area	No	401	95.5
	There is	19	4.5
	Total	420	100.0
Nervous system area	No	331	78.8
	There is	89	21.2
	Total	420	100.0
Burns area	No	408	97.1
	There is	12	2.9
	Total	420	100.0

TABLE 5 Distribution of areas of specific learning disabilities.

		n	%
SLD1	No	239	56.9%
	There is	181	43.1%
	Total	420	100.0%
SLD2	No	244	58.1%
	There is	176	41.9%
	Total	420	100.0%
SLD3	No	313	74.5%
	There is	107	25.5%
	Total	420	100.0%
SLD4	No	370	88.1%
	There is	50	11.9%
	Total	420	100.0%
SLD5	No	398	94.8%
	There is	22	5.2%
	Total	420	100.0%
SLD6	No	389	92.6%
	There is	31	7.4%
	Total	420	100.0%
SLD7	No	362	86.2%
	There is	58	13.8%
	Total	420	100.0%
SLD8	No	397	94.5%
	There is	23	5.5%
	Total	420	100.0%
SLD9	No	340	81.0%
	There is	80	19.0%
	Total	420	100.0%
SLD10	No	420	100.0%
	There is	0	0.0%
	Total	420	100.0%

SLD, specific learning disability.

Special Needs Levels according to disease areas are shown in Table 7.

4 Discussion

The problems faced by children with special needs are problems that not only children and their families but also the whole of society must face and find solutions to. The reports from SNRC health boards can help children and therefore their families to achieve some gains, which in turn can reduce and facilitate the problems. For this reason, studies like our study in which the reports of SNRC health boards are analyzed may be guiding for a solution (4, 7).

Of the 420 patients in our study, 58.3% were boys and 41.7% were girls. In parallel with our study, in many studies on SNRC, it was found that the rate of boys applying to the board was significantly higher than the rate of girls (1–4, 6–9). Some diseases that may cause special needs such as specific learning disorders, autism spectrum disorder, hemophilia, and Duchenne muscular dystrophy are more common in boys. Boys are also more likely to be exposed to negative environmental factors. A higher social awareness of behavioral and developmental disorders in boys may lead to easier identification of special needs. Therefore, gender should be considered an important factor in the identification and support of special needs.

Similar to our study in which the mean age was found to be 7.41 years, in many studies in Turkey in which SNRC reports were analyzed, it was found that the mean age of children who applied to the health board was 7–8 years (3, 7, 9, 10). Most families may not recognize the status of their children with mild special needs in the preschool period. At the age of 6–7, which is the age of school entry, families who suspect that their child may

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TABLE 6 Comparison of specific learning disabilities by gender.

			Male	Girl	Total	*p*
SLD1	No	Number	146	93	239	0.188
		Percentage	61.1%	38.9%	100.0%	
	There is	Number	99	82	181	
		Percentage	54.7%	45.3%	100.0%	
SLD2	No	Number	151	93	244	0.082
		Percentage	61.9%	38.1%	100.0%	
	There is	Number	94	82	176	
		Percentage	53.4%	46.6%	100.0%	
SLD3	No	Number	196	117	313	0.002
		Percentage	62.6%	37.4%	100.0%	
	There is	Number	49	58	107	
		Percentage	45.8%	54.2%	100.0%	
SLD4	No	Number	216	154	370	0.959
		Percentage	58.4%	41.6%	100.0%	
	There is	Number	29	21	50	
		Percentage	58.0%	42.0%	100.0%	
SLD5	No	Number	236	162	398	0.139
		Percentage	59.3%	40.7%	100.0%	
	There is	Number	9	13	22	
		Percentage	40.9%	59.1%	100.0%	
SLD6	No	Number	226	163	389	0.875
		Percentage	58.1%	41.9%	100.0%	
	There is	Number	19	12	31	
		Percentage	61.3%	38.7%	100.0%	
SLD7	No	Number	202	160	362	0.013
		Percentage	55.8%	44.2%	100.0%	
	There is	Number	43	15	58	
		Percentage	74.1%	25.9%	100.0%	
SLD8	No	Number	234	163	397	0.404
		Percentage	58.9%	41.1%	100.0%	
	There is	Number	11	12	23	
		Percentage	47.8%	52.2%	100.0%	
SLD9	No	Number	204	136	340	0.153
		Percentage	60.0%	40.0%	100.0%	
	There is	Number	41	39	80	
		Percentage	51.2%	48.8%	100.0%	

be an individual with special needs due to academic inadequacy take their children to the doctor. Sometimes the teacher may detect the difference in the child and direct the families to apply to the hospital. All these reasons explain why the average age of application to the SNRC health committee in most studies is around the age of seven. Therefore, for SNRC health boards to become more functional, arrangements should be planned to prioritize the needs of this age group. In addition, implementing projects to raise awareness among families in the preschool period and encouraging family physicians and family health workers to identify children with special needs may pave the way for early identification of children with special needs.

When the special needs levels of the children included in our study were examined, SCRN (90%–99%) was found with the highest rate of 47.1% by most studies in the literature (1, 6, 8, 9, 11). In the study conducted by Güller & Yaylacı, the highest rate of SEN was found to be 45.5% and the second highest rate of SCRN was found to be 27.7% (4). In line with some studies, when the distribution of special needs levels was examined in our study, it was observed that there was no significant difference between boys and girls (7–9). However, in some other studies, it was reported that there was a difference between male and female gender (1). The fact that the studies yielded different results reveals that the methods and criteria used in determining the levels of special needs should be re-evaluated.

When the distribution of the reasons for application was analyzed, no significant difference was found between boys and girls in our study and it was observed that the most common reason for application was to benefit from disability rights with 68.6% in boys and 64% in girls. In a study conducted by Kumbul et al. similar to our study, it was found that the most common reason for application was to benefit from disability rights (9). Unlike these results, in a study conducted by Kayhan & Öztürk, the most common reason for application was special education (52.4%) (8). The reason for this difference in the studies may be the difference in the perspective of special needs in the places where the studies were conducted, the fact that the primary need in children and families may vary according to sociocultural characteristics, the age distribution of the participants not overlapping, the different physical and environmental structure in the place of residence, and the lack of consensus among the physicians in the health committee on diagnostic criteria.

When the distribution of disease areas was examined in our study, it was seen that the highest rate was in the Cognitive Development Area with 36.2%, the second highest rate was in the Movement Development Area with 27.1%, and the lowest rates were in the Genitourinary System - Surgery Area with 0.2% and in the Hematology-Oncology Area with 0.2%. In the study conducted by Güller & Yaylacı, the highest rate of 44.7% was again in the Cognitive Development Field, while the second highest rate of 41.9% was in the Child and Adolescent Psychiatry Field (4). In the study conducted by Yıldız&Tarakçıoğlu, it was found that the highest rate was in the field of Child and Adolescent Psychiatry with 41.1% (2). These findings show that special needs can be seen in a wide range and at very different rates. Therefore, it is important to consider individual needs in the assessment and treatment of special needs. In addition, it is also important to closely monitor the Cognitive Developmental Domain.

In the present study, when the distribution of SLD areas was analyzed, SLD1 was found in 43.1%, SLD2 in 41.9%, SLD3 in 25.5%, SLD4 in 11.9%, SLD5 in 5.2%, SLD6 in 7.4%, SLD7 in 13.8%, SLD8 in 5.5%, and SLD9 in 19%. There were no children in the area of SLD10 (Table 5). This distribution shows that SLD1, SLD2, and SLD3 are at the forefront. This situation is important for both preventive medicine and diagnosis, treatment, and follow-up of children. In short, our study reveals that children should be followed up more closely in terms of certain needs. The way to achieve this is to educate both families and health professionals and to develop medical and administrative strategies that will lead to awareness in the community.

In the present study, no statistically significant relationship was found between boys and girls in terms of SLD1 ($p = 0.188$), SLD2 ($p = 0.082$), SLD4 ($p = 0.959$), SLD5 ($p = 0.139$), SLD6 ($p = 0.875$), SLD8 ($p = 0.404$) and SLD9 ($p = 0.153$). On the other hand, while

TABLE 7 Special needs levels by disease area (%).

Fields	No special requirements	Special needs (SCF): 20–39	Mild special needs: 40–49	Moderate special needs: %50–59	Advanced special needs: 60–69	Very Severe special needs: 70–79 (Considered to be Severely Disabled)	Significant special needs (SSN): 80–89 (Considered to be Severely Disabled)	Special condition requirement (SCRR): 90–99 (Considered to be Severely Disabled)
Cognitive development area	4.0%	28.0%	4.0%	0.0%	0.0%	4.0%	0.0%	60.0%
Child and adolescent psychiatry	3.3%	8.3%	3.3%	6.7%	1.7%	5.0%	3.3%	68.3%
Speech-language-communication development	0.0%	19.7%	2.6%	14.5%	1.3%	6.6%	3.9%	51.3%
Endocrine system area	0.0%	33.9%	1.6%	3.2%	0.0%	1.6%	4.8%	54.8%
Genitourinary system - surgical area	0.0%	60.6%	3.0%	3.0%	3.0%	0.0%	3.0%	27.3%
Visual function area	0.0%	20.0%	0.0%	0.0%	3.3%	0.0%	13.3%	63.3%
Movement development area	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	100.0%
Hematology-oncology area	0.0%	35.7%	7.1%	3.6%	7.1%	3.6%	0.0%	42.9%
Hearing function – ear, nose, and throat area	0.0%	25.4%	2.6%	8.8%	2.6%	2.6%	5.3%	52.6%
Hereditary-natal diseases area	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	100.0%
Metabolism area	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	100.0%
Nephrology area	0.0%	0.0%	0.0%	0.0%	0.0%	0.0%	50.0%	50.0%
Digestive system area	5.3%	15.8%	0.0%	10.5%	0.0%	5.3%	15.8%	47.4%
Nervous system area	3.4%	18.0%	2.2%	7.9%	3.4%	5.6%	4.5%	55.1%
Burns area	0.0%	16.7%	8.3%	8.3%	8.3%	16.7%	8.3%	33.3%

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SLG3 was higher in girls (54.2%), SLG7 was higher in boys (74.1%) (Table 6). This result reveals that boys and girls should be evaluated separately in the evaluation of SNRC in planning for both prevention treatment and rehabilitation.

In the present study, when the levels of special needs according to the disease areas were analyzed, the highest rate was determined as SCRN in 13 areas except for the field of Nephrology and Genitourinary Systems. In the field of nephrology, the rates were equally shared between SCRN and SSPD at 50%, while in the field of Genitourinary system surgery, the highest rate was found to be SEN with 60.6% (Table 7) In a study conducted by Kumbul et al. in Isparta in 2020, it was found that a different need had a higher rate in almost every field, which was very different from our study (9). These differences may be due to the difference in the geographical region where the study was conducted, as well as the change in the profile of patients admitted to the hospital due to the impact of the COVID-19 pandemic and the change in the expectations of families to apply to the health committee. The identification and management of special needs can be influenced by many factors such as geographical healthcare infrastructure and sociocultural dynamics. This also illustrates the difficulty of standardizing special needs for adults and children. In other words, the most important outcome of our study is that individualized health services are vital.

Many activities and programs are recommended for children and young people with special needs to be more active in social life and to increase the self-confidence of themselves and their families (12). However, the most important point here is to pave the way for children to be diagnosed correctly and to benefit from their legal rights in the maximum way. The primary way to achieve this is to develop strategies to ensure that the SNRC health boards are structured in a faster, more objective, and more inclusive manner through future studies.

The biggest limitation of the study is that it is not known how many of the families of the children included in the study objected to the report and whether the results of the objected reports changed. Another limitation of the study is that the physicians in the health committee changed within 5 years, and accordingly, the patients included in the study may have been evaluated by physicians with different approaches. In addition, since Batman, where the study was conducted, is located in the Southeastern Anatolia region of Turkey, the results may vary too much to be generalized to other provinces with significant geographical and cultural differences. The advantageous aspect of the study is that it covers the entire five-year period from the first day of the regulation of the SNRC.

5 Conclusion

The study shows that SNRC health board reports can mitigate crucial problems by providing important benefits for children and their families. In this context, analyzing SNRC reports can serve as a guide in determining solutions. The findings of our study revealed that gender is an important factor in the identification and support of special needs, and the age of presentation is usually 7–8 years. In addition, it is of great importance to develop strategies to raise awareness in the community and to train health professionals. Further research and continuous monitoring are crucial for improving outcomes and support systems for these children and their families.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Clinical Ethics Committee of the Batman Training and Research Hospital [approval number: No. (25.01.2024)-375]. The studies were conducted in accordance with the local legislation and institutional requirements.

Author contributions

SC: Writing – original draft, Data curation, Methodology, Investigation. MP: Data curation, Formal Analysis, Methodology, Writing – review & editing. EK: Writing – review & editing, Data curation. EO: Data curation, Resources, Writing – review & editing.

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Conflict of interest

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

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Opportunities for meaningful inclusion: experience of individuals with intellectual and developmental disabilities with research

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Background: Individuals with intellectual and developmental disabilities (IDD) face numerous health disparities, particularly in rural communities. However, they are rarely included in the research process to address these challenges as co-researchers. Little is known about the experience of how individuals with disabilities participate as co-researchers, or the barriers they face.
Objective: The current study explores the experiences of individuals with IDD as co-researchers through discussions with individuals with IDD themselves, those who support them, and disability researchers.
Method: Data were collected through focus groups with individuals with IDD, individuals who support those with IDD, and disability researchers. Each group was asked about their journey through the research process, from beginning to end. Data were analyzed thematically by two independent coders.
Results: While all groups viewed the inclusion of individuals with disabilities as co-researchers as valuable, many barriers still prevented this population from fully participating in the research process. Individuals with IDD viewed research positively, especially when the topics were personally relevant. However, many thought research was intimidating and wanted additional support. Support providers expressed that the people they support have lots to contribute to research and felt empowered when participating. Disability researchers discussed many barriers to include individuals with IDD as co-researchers, including limited time, resources, and inflexibility of research processes. Researchers felt they could use more experience working with individuals with disabilities as co-researchers to integrate these individuals into all aspects of the process.
Discussion: There is broad interest in including those with IDD as co-research, but many barriers remain. Full inclusion can be supported by developing a welcoming and accessible environment. Researchers may need institutional support and training to pursue inclusive IDD research. Asking individuals with IDD for their expertise, develop topics of research that those with IDD can relate to, and involving support providers may be helpful. Developing innovative strategies to support inclusion is needed from all groups.

KEYWORDS

intellectual and/or developmental disabilities (IDD), journey mapping, research participation, co-researchers, engaged research

Introduction

Individuals with intellectual and/or developmental disabilities (IDD) face persistent healthcare disparities. For instance, those with IDD have poorer overall health outcomes, such as an increased likelihood for obesity and physical inactivity, and a decreased likelihood to receive screenings for cervical, breast, and prostate cancer (1). This population also has increased use of emergency services related to social determinants of health (2). Those with IDD also have higher rates of chronic physical and mental health conditions, emergency department use and 30-day readmission rates (3). Adults with IDD also have significant gaps in their sexual health knowledge (4), have higher rates of interpersonal violence (5), and have higher rates of adverse childhood events and associated long-term negative health consequences (6). Overall, those with IDD struggle with numerous health issues related to access, knowledge, communication and quality (7).

Unfortunately, these negative health outcomes are exacerbated for those with IDD who live in rural settings (8). This is not unexpected given that those in rural and frontier communities experience lower life expectancy and poorer overall health status (9). Rural populations have reliably lower County Health Rankings in areas such as behavioral health, morbidity, and access to clinical care (10). County Health Rankings measure the population health of counties based on interconnected factors related to the policies and procedures of local, state, and federal governments with health factors and health outcomes such as length and quality of life (11). This is strongly influenced by individual and environmental characteristics, including higher rates of risky health behaviors, limited financial resources, socioeconomic differences, limited access to health care, poor health care quality, limitations in infrastructure, insurance deficiencies and a weak public health policy environment (12, 13). These disparities affect all rural patient demographic groups (14) and disability status only compounds these challenges. Therefore, rural residents with IDD are far more likely to face significant health disparities.

Health research is typically a critical step in determining how to remediate highly prevalent health disparities, such as those listed above. Indeed, this has been the cornerstone of public health research for over 100 years. More recently, there is a growing interest in how to bridge the gap between researchers and individuals in impacted communities. New models of community engaged research (15) provide a path to conduct research *with* the community, rather than *on* the community and may produce more locally relevant outcomes. Community-based participatory research (CBPR) is a type of engaged research that prioritizes the inclusion of all stakeholders in all phases of the research process. CBPR research is a collaborative process that includes community members as *co-researchers* in the development of the research questions, methods, data collection, data analysis, dissemination, and determining how the findings will be used to benefit the community (16). A growing body of evidence suggests that programs that draw upon the experience of community members to identify health priorities that matter most to them are more likely to build healthcare capacity and quality in rural communities (9) and for those with IDD (17). The use of CBPR is of particular interest with IDD and rural communities because it is more effective in developing solutions that fit unique local circumstances and, therefore, maybe more likely to lead to sustainable positive change. This is particularly important for extremely rural and underserved communities that have very little capacity to try new programs. Therefore, it is it is important to enhance collaboration among all stakeholders that align with the common interests of the community and professionals (18) and tailor this method of research to better conduct research with individuals with IDD and those in rural communities.

Unfortunately, little is known about the experience of those with IDD in health research, nor how researchers who conduct health research related to IDD include those with IDD in their work. The research needed to remediate these disparities has traditionally excluded those with IDD from the process of research (17, 19). In fact, there has been a long-standing tension between researchers and individuals with IDD, further complicating participation in research by individuals with disabilities (20). Further, given the vastly different healthcare challenges in rural areas as compared to urban areas, solutions that are developed for urban centers are not likely to be effective in rural communities (21). This includes economic factors, cultural and social differences, lack of public health education, political factors and the sheer isolation of living in remote areas all conspire to impede rural Americans in their struggle to lead healthy lives (14). Therefore, solutions that are designed to meet the unique needs of those with IDD living in rural America are desperately needed.

To encourage inclusion of those with IDD as co-researchers, it is essential to understand the experiences individuals with IDD have already had in research, as well as what motivates these individuals to help with research and what barriers they experience. One process that has been used to understand the experiences of individuals in various settings is called journey mapping. Journey mapping is a process commonly employed by healthcare administrators in order to understand care management from the patient's perspective (22). Instead of viewing the patient experience through the healthcare system perspective that may prioritize specific events that are salient to the healthcare system (e.g., patient seeks help, care provider makes diagnosis, patient receives treatment), journey mapping focuses on every activity or event between the patient and their healthcare system that shapes the overall experience (23, 24). For example, journey mapping includes prescription or appointment reminders as well as direct contact with providers in order to identify any gaps in the patient's experience (23). Therefore, the journey mapping process focuses on the whole system, and prioritizes the respondent's point of view.

Journey mapping can be tailored to any population or situation by exploring key experiences, including successes and challenges, that populations face throughout their participation in specific scenarios. For example, in CBPR, co-researchers are expected to contribute to all steps of the research project, including

formulation research questions, data collection, data analysis, and dissemination of project results. Utilizing journey mapping as a research tool would require researchers to ask participants about their experiences with all aspects of the research process to further understand barriers to full participation of co-researchers. This tool can then be further modified to focus on the experiences of specific populations, such as the experiences that individuals with IDD have had contributing to research. Further, journey mapping becomes particularly powerful when the experiences of multiple stakeholder groups are considered, compared and contrasted. This allows for a better understanding of the entire social system at a functional level, and uncovers novel areas for intervention.

Therefore, the current study uses journey mapping, adapted to the process of inclusive IDD research to explore the experiences of (1) individuals with IDD, (2) their support providers and (3) disability researchers in the inclusion of individuals with IDD in the research process. Discussions with these three groups will help to identify previous experiences with research, as well as any successes and challenges in integrating individuals with IDD as co-researchers. Further, the current study will provide guidance to multiple stakeholder groups on how to address barriers to the inclusive IDD research. The involvement of individuals with disabilities as co-researchers will allow for more inclusive research, and hopefully lead to valuable research outcomes for individuals with IDD.

Method

Participants

Participants fell into three groups: (1) individuals with IDD, (2) individuals who support someone with an IDD (e.g., family members, case managers, direct support providers, etc.), and (3) researchers who engage in research related to disability. Participants were required to be 18 years of age or older. Anyone who identified as someone with an IDD was included in the IDD group. Proof of a formal diagnosis was not required. Individuals with IDD who were their own guardian or had a guardian were permitted to participate. All participants received $50 for their time, given as a gift card.

A total of 9 individuals with IDD, 5 researchers, and 4 people who support someone with an IDD participated in the focus groups. All participants worked and/or resided in Wyoming. Of the participants from the supports group, 1 participant was a family member, 1 participant worked with individuals with IDD, and 2 participants had a dual role of family member and someone who worked with individuals with IDD. All 5 researchers conducted research related to disabilities. Groups were recruited in two ways. The IDD group and supports group were recruited at the Wyoming Developmental Disabilities Conference held in June 2023. All attendees of the conference were invited to attend a pre-conference workshop that explained what research was and the goals of the current research project. Interested participants completed the eligibility and consent process during this workshop. Researchers were recruited through outreach to the University of Wyoming faculty email list, the University of Wyoming College of Health Sciences faculty email list, and the Equality State Research Network (ESRN). Focus groups for researchers were held via Zoom.

This research was determined to be a low-risk study; however, because of potential participants with IDD are part of a protected class, institutional review board (IRB) regulations stipulated that individuals with IDD must demonstrate understanding of the purpose of the research, their rights, potential risks and benefits prior to participating. The individual collecting consent would ensure understanding by asking the individual to repeat back key information about the study, including, but not limited to the purpose of the study, their right to choose to participate or not, that they could stop participating at any time, and that they could choose not to answer any of the questions. This understanding check was conducted prior to consent, and only one individual was unable to complete the comprehension check. If the participant had a legal guardian, a researcher contacted the guardian via email or phone call to obtain approval to discuss the project with the participant prior to administration of the capacity assessment. If the participant demonstrated a capacity to consent and was their own guardian, a written consent form was presented to the participant. If the participant had a legal guardian, the guardian received a consent form, and the participant received an assent form.

Procedure

Three focus groups with self-advocates and care providers were conducted at the 2023 Wyoming Developmental Disabilities Conference. Researchers from the Wyoming Institute for Disabilities (WIND) held two focus groups with individuals with IDD and one focus group with individuals who support someone with IDD. Two focus groups with IDD researchers were held via Zoom. Focus groups lasted around 60 min. Participants were asked what experiences they, or the individuals they supported, had with IDD research. Participants were encouraged to share successes, difficulties, barriers, and thoughts about including individuals with disabilities in the research process, including creating research questions, collecting and analyzing data, and disseminating results. Interviewers were all staff and faculty of the WIND who had experience working with individuals with IDD.

Measures

Data were collected using a structured interview guide. The guide was built around the core principles of CBPR (16) to capture experiences with each of the phases of the research process. Questions were specifically tailored for IDD participants, their support providers, and researchers resulting in eight open-ended questions, with follow-up questions (see Supplemental Materials). These questions focused on general experience with research as well as specific questions about developing research

questions, data collection, data analysis, and dissemination. Follow-up probes were used to gather more detailed information and to refocus the conversation, as needed. Focus group questions were developed based on the Levy method of journey mapping by determining touchpoints, timeline, external influences, internal influences, and barriers to participation in research (22). To further tailor this tool, researchers discussed experiences relevant to the participant groups in this study, rather than focusing on healthcare systems and their patients as the tool was originally developed for.

Data analysis

Following the collection of data, transcripts were generated using an online AI-based transcription platform. Prior to analysis, a research associate listened to each interview along with the transcript to correct errors. Transcripts were then analyzed using a thematic analysis approach to determine reoccurring themes across participants and groups based on the shared experiences of participants (25). A thematic analysis approach was chosen to gain in-depth insight into the experiences of individuals with IDD, their supports, and IDD researchers, and explore the similarities and differences of each participant and group. Furthermore, thematic analysis aligned with the goals of the current study, which were to gather the experiences of each group in their own words. Collecting and analyzing data that corresponded directly to the thoughts and experiences of each group allowed for an authentic representation of the journey these participants have had in research.

Several steps were taken to ensure credibility in the findings. The research team comprised of an interdisciplinary team with two faculty members and two staff members. Faculty included CH (public health and qualitative methods) and EM (social psychology and epidemiology). Staff were AD (clinical psychology) and TB (public health and administration). One of the team members identifies as a family member of individuals with IDD. This diversity of perspectives provided a unique lens to this study data.

We also made attempts to enhance the credibility of findings by taking a team approach to data analysis. Two independent coders (AD, TB) listened to each recording before comparing common themes. Coding was completed manually by each coder. To begin this process, each coder read the transcripts from each focus group to become familiar with the data in general. In the initial coding phase, coders used an inductive coding (26) approach called line by line coding to ensure all data was thoroughly analyzed. As coders read through each comment made by participants, they would code lines with descriptors that synthesized the general meaning of the comments. When this process was complete, coders organized their codes into common themes. Following this process, the coders met to compare the themes that emerged and read through transcripts while marking common themes discussed by each group. When coders disagreed on themes, they discussed what evidence was placed under that theme to determine if there was a match to another theme that was categorically similar. Additionally, if one coder split themes into two categories, while the other coder combined those themes, they discussed whether the themes were distinct and came to a consensus on whether to split or combine themes. If a theme arose that only one coder recognized, coders would review the evidence behind that theme and decide together if it should be included in the final list of themes, integrated into another theme, or disregarded. The coders then discussed emerging themes that were in common and developed a final list of themes. These themes were then discussed amongst the entire author group to develop consensus. Several themes were identified for each of the groups, which are outlined below.

Results

Individuals with IDD

Four main themes emerged from the focus groups held with individuals with IDD: (1) Positive View of Research, (2) Personal Topics, (3) Research is Intimidating and Inaccessible, and (4) Increasing Comfort in Research. While participants had an overall positive view of research, most had very little experience with any form of research, and those that did, only as a research participant (i.e., as a subject). None had experience as a co-researcher or providing any supportive role to a research team.

Positive view of research

Overall, participants indicated that they had a positive view of research. As noted above, none of the participants had ever served as a co-researcher, but many had been research participants. This mostly included answering questionnaires and interviews. However, most participants with IDD spoke of valuing the ability to be a part of something important and helping to solve problems that are important to them. When talking about helping to form research questions, one participant expressed interest in being involved in creating research questions, noting the potential for unique contributions as a result of having an IDD: "*My brain is different, okay my brain. And my brain is different.*" This comment was made in the context of being able to offer a unique perspective as a result of these differences.

Many comments suggested a deep appreciation of participating in research in any way. Several comments focused on how it allows them to use their voices in ways that they usually were not asked to. "*I think it kind of forces you to think creatively about different ways to solve the issue or find solutions to the theme you're researching. Because maybe the typical route to it isn't always the route that works.*" This further suggests an awareness that the unique contributions of people with IDD can help enhance research.

When participants were asked if they were interested in participating in similar research in the future, most participants enthusiastically said they would. When the group was asked if they liked helping with research, one participant stated, "*Yeah. I like it, I like it here. People helped me out. To talk about*

research. I liked it here." Another participant expanded on this by saying,

> For me what I like about it is, I know that it helps to educate people and make sure that they are getting the services that they need, not only for myself, but it helps educate … people about disabilities, I really feel like it's important to educate people on the needs and disabilities and I've always thought that since I have a voice, why not be a voice for other people that don't have a voice. And really get that education out there. And I felt like surveys are a great way to advance Disability Services. And anytime I could be a part of that. I feel like it's very important. So, I take it very seriously. And I automatically, whenever I can, I automatically always participate in surveys whenever possible.

Personal topics

When asked about what topics they hoped to see in research, and what research questions they would like to develop, participants focused on topics that had a personal connection to their lives. Perhaps unsurprisingly, there were interests that extended well beyond healthcare research. For instance, participants discussed health and wellbeing, relationships, jobs, community accessibility, and disability policy. Each of these topics was discussed in relation to personal experiences in the participant's own life. For example, some participants were interested in research regarding social security benefits and marriage, while others were interested in research related to jobs for individuals with IDD.

Each of these topics had a personal impact on the participant that made the comment, and respondents usually could provide clear explanations of the personal value to their life. For instance, several participants had specific interest in health, with one noting an interest in understanding diabetes better: "*Like, figure a way, like, to be healthy*". Non-health related topics of interest often related to social structures, healthcare systems or policies that had significant impact on their lives. One respondent noted a strong interest in potential problems related to an upcoming marriage and how it could impact his social security and other benefits: "*… whenever we get married, I'm going to be losing all of my SSI benefits … And we're trying to, and I'm trying to figure out why that is or what the reason is behind it.*" Several other comments focused on health care access, especially through Medicaid. The common theme among these topics is that they are meaningful to the participants with IDD and the results could have an impact on their lives.

Research is intimidating and inaccessible

One of the major barriers preventing those with IDD from participating in research was a concern that research is too intimidating and hard to understand. This included several unique facets, but focused predominantly on not understanding the research process, and inaccessibility of materials. For example, most participants indicated that they would not know what to do if they were asked to help with research. This seemed to relate to a general lack of knowledge about the research process. However, most suggested that they would be willing to learn. For instance, several participants stated that they would need to start with smaller tasks to help build their confidence before they were able to feel comfortable helping with a research team. When talking about getting involved with data collection and interviewing research participants, one participant talked about starting with smaller, less intimidating tasks:

> I'd start with people I know first and then slowly work my way up to people I don't know. Because I'm one of those that like feed off of… encouragement. Like, if I know that…I could get somebody I know to fill out a survey and I'm like, okay, I kind of got this.

Other participants talked about inaccessibility of materials. Several mentioned how research can be hard to understand, especially when words are used that they do not understand, or instructions are not repeated. During a discussion about not knowing what research means, one participant stated, "*Yeah. Especially when you use big fancy words*". Another participant agreed that understanding what was being discussed could be difficult, and they would need things repeated to them in order to understand, "*…please repeat it. Again, and hard to understand. hard, hard to understand. Like, like, you know what that means?*".

Difficulty with traveling to where the research is conducted was also commonly discussed. One participant noted "It's kinda..Well research..What's this? It's really hard for me, it's hard for me because I'm doing this on my own. I left [Wyoming town] last night and then come here [Wyoming Town – about 150 miles away]." This respondent noted that this amount of travel was particularly difficult and made it hard to participate in research, which was echoed by others.

These concerns also led to participants also worried about being rejected by researcher and fear of social situations related to research such talking to people they did not know when collecting data. While discussing their fears of helping with data collection, a participant said, "*Especially walking up to a complete stranger. It's like, this is a con. Yeah*". Overall, this suggests that research is both intimidating and inaccessible at times for individuals with IDD, but individuals are willing to learn and grow their skills.

Increasing comfort in research

Despite their concerns about joining a research team, several participants had ideas of how they could be more successful as a co-researcher. When discussing how to get involved with research indicated that they would feel more comfortable if they were able to participate with someone who would provide support to support increased engagement in research. This support related to several technical components of participation such as scheduling, transportation, and use of technology to complete surveys. Respondents noted that they thought a support provider could help them manage the research process better and ease any discomfort individuals with disabilities may have.

Interestingly, this was not exclusively a professional support provider (e.g., DPS), but could include people from a variety of formal or informal roles. For instance, when participants were asked who they would like to be present to help with research, participants had many ideas including, "*Maybe an older, an older person in your life*", "*a mentor, or like a coach, or a teacher*", "*family member, a case manager*", "*provider*", and "*friends that you talk to*".

Indeed, many respondents pointed out that building relationships with peers with similar research interests may help them engage in the research process. When talking about the conference where the focus groups were held and avenues where individuals with IDD could participate in research, participants generally agreed noting:

> Maybe like so if you all wanted to help us actually collect data, this might be a place where we maybe we train you up beforehand, and maybe talk with job coaches, and case managers and say, "We want you guys to help with this." And we can even make it a bit of a your job, right? pay a little bit of money. And you could help us collect data.

Generally, many participants thought that working in a group of people with similar experiences and interests would increase their comfort to talk about the research that they are passionate about. When discussing how to get research started a participant stated:

> And then you guys can all. You guys, all of us can just all like group together and not essentially peer pressure. But peer pressure these people into helping us answer the questions and doing the research that we [want].

Support providers for individuals with IDD

Like the IDD group, most support provider participants reported their clients and/or family members had participated in some sort of research in the past but had not been involved as a co-researcher. These respondents generally regarded research as a positive experience for their clients and loved-ones. Five themes emerged from the support providers: Enthusiasm to Participate, Empowerment, Inclusion, Safe Environment, and Translatable Skills.

Enthusiasm to participate

A very strong theme emerged from this group indicating a belief that their clients would be interested in participating as co-researchers. Overall, there was high enthusiasm for participation and each person stated that if the people they support were given the opportunity to help with research, they would take it. One participant stated, "*Like some of my participants, they would be here in a second telling you how much they love you. And they cannot wait and give you the answers that you're looking for*". Another participant mirrored these thoughts when it came to a family member, "*…once she gets involved, she's more than ready to give information about what her life or her experience has been*". A third participant agreed that some of her clients would be interested in participating in research:

> I can think of three or four [clients] right now that they would love that. They would love to be able to say, you know, these are the types of conversations that I want to have with somebody and so maybe somebody else would too.

This group made it clear that the people they support want to be heard if people are willing to ask them about their experiences.

Empowerment

Participants from the provider group discussed that participating in research has allowed the people they support to feel empowered. These respondents noted that when individuals with disabilities are asked to participate in research, they are excited to talk about their experiences and have feel like they have some control over the research that they are helping with. A family member of an individual with IDD said, "*I think it's a sense of inclusion. And accomplishment that someone's listening to me.*" Additionally, participants noted that having an opportunity to talk about their experiences in a group setting is empowering because there are often like-minded individuals who have had similar experiences listening. One support provider stated:

> I mean, at least in what I've seen from my participants, even if we … break down … the survey into like a smaller degree, and not just like, in this sort of capacity. I feel like there's always like a sense of empowerment, because typically you're in a group setting, there is the capacity of being around people who can relate.

Further, participants commented that using research as a platform to advocate for oneself and for others with disabilities is very impactful for the people they support. They like that their contribution to research could help other people with IDD. One participant stated, "*that's where I think my clients would be like, yes, we need this information so that other people, you know them included, have that benefit*". The empowerment from participating in research makes it more likely for individuals to seek out research, as well as other empowering activities, in the future.

Inclusion

Participants in this group expressed that the people they support want to be asked to participate in research and they want to be listened to throughout the whole process. They indicated that this would promote a sense of empowerment and make individuals with IDD feel respected. Several respondents also noted that in previous experiences with medical appointments and research experiences, individuals with IDD

were usually not asked questions directly. One participant reflected on this:

> I think one thing that happens quite often, and whether it's an interview or a questionnaire, those people asking the questions, if you are with your child, or your person you're advocating for, or you're, if you're a respite worker, they always tend to ask the adult or the normal physical person instead. And so many times I said, don't ask me, ask her… And so,… I find that happens quite often in this kind of informational part where they start asking questions, they need to ask the individual and see if they are capable or not. But they jumped to conclusions.

Additionally, participants in this group emphasized a need for communication throughout each research step, including improved communication during data collection and dissemination of results. One participated noted that in a research study, they stopped receiving communication from the researchers, "*Yeah, you expect a kind of a follow up or reason, or maybe we've terminated this information or research that just dropped with no reason or rhyme*". Many participants noted that the people they support never received information about the results of the studies they participated in, leaving them confused about the outcome of the study and how it impacted individuals with disabilities. One participant stated:

> Yeah, you know, where are the results? Yeah, we did this stuff. And I never got anything. You know, my daughter did this whole thing. And it was a couple of years. I have no idea what their results were… How did it help them? What did they get out of it? I have no clue. And I have no way to get that information. So, I know that, you know, my daughter doesn't have that information. My clients don't have that information.

Safe environment

When discussing the ideal circumstances for individuals with disabilities to participate in research, many support providers discussed the need for a safe environment. This group noted that individuals with disabilities need to know that they will not be judged for what they say and that they are in an environment where they are allowed to freely express themselves. One example of a safe environment was the conference where the focus groups were held. When asked if the conference felt like a safe environment for individuals to express themselves, one support person said:

> 100% Because they are with their like-minded peers, they are with new peers. There's new things, and then you've got this amazing speaker who's just making you feel all kinds of happiness? Absolutely. Absolutely. Yes. It's more an open environment.

Respondents noted that this type of open environment develops a sense of comfort for individuals with disabilities and creates an opportunity in which they can formulate their own stories to share with researchers and their peers. Participants stated that the people they support are often left out of conversations or feel they are in environments that are too restrictive for them to share their thoughts. When discussing another speaker at the conference that had talked about his experiences not being included due to his disability, a family member shared:

> Yeah, the one individual today, he had such a powerful message. But if you looked at him, you would think he was angry. But he wasn't. He was so happy to say what he said. And it's like I just applauded him for. And I don't know, if you would put him in a more open environment, people would have been maybe offended, by the way he approached that, just the way he looked. And it doesn't matter how he looked. His message was very important. Yeah, so. But it was good that he had the courage to say what he wanted to say, and everybody listened.

Translatable skills

Most participants from this group reported that the people they support did not have any formal experience in helping with any aspects of the research process. However, many participants discussed skills and experiences that they thought would be beneficial in supporting research projects. Specifically, participants discussed a range of experiences and skills related to critical thinking, resourcefulness, and problem-solving. For instance, when asked if the people they supported had ever interviewed participants, one support person stated the people they supported participated in the hiring process at their group home and asked potential staff questions during the interview process, "*It's just interviews that they will give somebody who we'll hire. Alright, so they were doing the interviewing on if they're going to hire them or not.*" Additionally, when participants were asked if the individuals they supported had ever assisted with planning a study, one participant stated that their daughter had organized a birthday party.

> …when my daughter was going through transition classes, and you know, past 18, but not 21 yet. You know, they would sit down, and the group would have a discussion on a, okay, well such and such birthdays coming up. So, we're gonna have, we're gonna do like a lunch. We're gonna go to the park and have a picnic. And so, then she would automatically take the lead and get everybody's information. And then she would compile that and talk about it. And, okay, well, this is what most people want. And so, this is what we're going to do. And now we need to make a budget, so we know get those things. And then we have to put those things together. So, she would direct. And I think she would take that information and kind of figure out what was going

> to work in the process. So yeah, it's the same. Like, it's the same process.

Other examples of experiences with translatable skills included participating in science fairs in school, participating in speech and debate clubs, propagating plants, and counting the number of new animals added to a family's goat herd. In general, this group thought that there were many skills that people with IDD can use to support research projects.

Disability researchers

Like the previous two groups, the group of researchers noted that those with disabilities rarely ever participate in research as co-researchers. This group noted several obstacles that discouraged the inclusion of individuals with IDD in research. In discussions about these obstacles, four prominent themes emerged: Structural Barriers, Comfort/Training, Advisory Boards, and Disconnect Between Academic Processes and What Individuals with IDD Need.

Structural barriers

The most prominent theme among disability researchers were the structural barriers that they faced in including individuals with disabilities in the research process. The most common barriers discussed were resources (i.e., money, childcare, translators) and time. In relation to resources, researchers discussed how it was difficult to find funding to compensate participants and research assistants. Additionally, finding funding to help co-researchers with disabilities attend conferences was a barrier. One researcher talked about the cost of attending conferences:

> So like, like I'm planning on going to [conference] in [month], well that's in [state]. So it's like, … actually a requirement of the [grant], which is what I want too. What I'm presenting on, is that I cannot use those funds to pay for a conference. So, I can't pay for a participant to come with me to go. So, then it's like, well, how do I get them there? And it's very costly, too.

During discussions about time, researchers talked about how much of a time commitment CBPR can be. It can be difficult to find individuals, including those with IDD, who are willing to commit a significant amount of time to a project. The time commitment can be intimidating to both researchers and individuals with IDD who may otherwise be interested in helping with research. When talking about time, a researcher commented:

> …and if we're talking about like, participatory research, like that's a big time investment. It's not just like, let me interview you and then I'll give you your incentive. That's one thing but if we're talking about, like, being a part of this whole thing, and we really want you invested, Yeah, it's great when people want to be involved and people volunteer and stuff like that, but I also think people should be compensated for their time.

Related barriers discussed include a lack of flexibility in research plans, such as having inaccessible meeting times for community members, and the inaccessibility of many research tools and spaces. One researcher reflected on the accessibility of the research tools they were using and how they learned from these unexpected accessibility challenges:

> I mean, I think it's always challenging, especially when you're researchers that don't presently have a disability. Because there's always things that we don't anticipate, that we didn't plan for, right? And especially in terms of accessibility. Like, for instance, the [name of project] that we did this past year, we brought in these cameras that my colleague had and they were just these little point and shoot cameras, but they were too small for the people and they we didn't have tripods and some people's hands shook, you know, and so we just like really learned a lot about, like, okay, we need to go out and we need to get some nail polish to mark certain buttons for them. We need to get tripods we need to get bigger cameras, maybe some iPads, just to make that more accessible.

Comfort/training

Although a less common theme, a few researchers discussed a lack of comfort in including individuals with IDD, for both themselves as researchers and for co-researchers with IDD. For some researchers, they were cognizant that they lack the training they need to know how to effectively include individuals with IDD in the research process. These respondents also talked about a lack of experience in conducting research on topics that individuals with IDD may be interested in. For example, one researcher noted a personal lack of technical training to approach questions about larger, census level research questions that some individuals in the community had posed:

> …And so it seems to me that the folks in the community have ideas about what needs to be done. And for me, a barrier is that I don't have any training doing that at all. And some of the ideas that folks have shared with me and these are caregivers and individuals with [disorder] are like census level type of questions, which again is like seemingly so out of my wheelhouse.

This group also discussed a potential lack of training for co-researchers with IDD as well. Specifically, many researchers discussed that many individuals with IDD, although interested in helping, may not have a background in research methods to be effective. Research worried that this could become more complex in later stages of the research process, such as analyzing complex data. While talking about data analysis, one researcher noted:

> I think is harder for quantitative analysis, maybe qualitative analysis to come out with a theme of different comments,

> I think that'd be very valuable. But quantitatively, I think it'd be quite hard to do the stats do the math, that would be harder.

This group expressed concern that researchers must train these new co-researchers, starting with basic research methods. One researcher reflected on a previous research study where individuals with IDD were involved:

> One was that I think, kind of a problem that you have, when you're working with people who don't engage with research very often is just like, explaining what is a research question. And, and then getting, so, it's like, basically, you have to teach intro to research, in order to, to involve… involve anyone into in to a research project. And, so that was true of people with intellectual and developmental disabilities, too. So, and that really went back to very basics, we had to ask, like, we had to talk about, like, what is a question? What makes an interesting question? What's a research question? What makes an interesting research question?

Advisory boards

Several researchers reported utilizing advisory boards comprised of individuals with IDD in previous studies but worried about the degree to which this was meaningful or impactful. These advisory boards were largely used as sounding boards in the beginning of studies to ensure that study procedures and the questions that were asked would be appropriate for the target population. In some cases, advisory boards gave some input in the interpretation of data or were given reports of outcomes. One researcher noted:

> We brought the results back to the advisory board, and kind of told them what it meant. And, you know, because it's statistical analysis, yada, yada, yada. And what we were finding and asked them sort of like, well, what do you think about this? How does this make sense to you, like questions like that, but and I think that that process is important, but I don't know how, how meaningful it is either. Because I don't think like we did that because it was part of our process. And a step like we were we were dedicated to we… were committed to including the advisory board in every part of the process, and also including them on publications. But I don't know if we actually included any of that input in our analysis, it was still more just, this is what the statistical analysis says, and this is how the researchers are interpreting it.

Similarly, most researchers reported their advisory boards did not participate in other vital research processes, such as data collection, data analysis, and dissemination. Researchers attributed these deficits in participation to the barriers discussed above, as well as a lack of interest in other parts of the research process. When discussing previous experiences with advisory boards, one researcher stated:

> For me with my community advisory council, I did ask if any of them wanted to help as far as interviewing. So, I gave them the option because I didn't want them to feel obligated to have to. And I certainly didn't want them to feel uncomfortable doing so. And none of them expressed an interest. So, I didn't, I didn't push it, because I was grateful for the assistance in creating the questions.

Further, several researchers commented that they believed they should include individuals with IDD in more aspects of the research process but had not done so due to the numerous barriers noted. One participant stated:

> So, I think in that sense, yeah, paying people… maybe actively seeking out research assistants with disabilities that are members of the community that you're working with? Yeah, yeah. I don't see why it can't be done. And it probably should be done more often, I would say too.

Another researcher mirrored those thoughts while reflecting on their own research and how they had not asked people with IDD for their input:

> And now I'm wondering, because as like, from my clinical training, I have all these ideas about, you know, what makes the communication environment hard, and what we can do to reduce the load of that communication environment and make it more successful. And now, I just think maybe I should ask [community members]. You know, because I have all of these ideas. I know it's shocking. I'm really glad we're having this. You know, I think it's easy to get lost for me in what we already know. But we haven't… I haven't really revisited that. And maybe there's contexts that these folks are experiencing whether you know, maybe they're outside a lot on their property, are these contexts that I don't even know.

Disconnect between academic processes and what individuals with disabilities need

One aspect that many researchers commented on was how the typical functioning of academia does not fit well with modified processes that would benefit individuals with IDD. For instance, participants stated that research requires rigorous methodology and changing procedures to be more accessible for co-researchers with IDD may be difficult. One researcher noted, "*it's hard, because it just in the science world we're so strict about how you control everything, is getting, yes, you have to do things by the literature. And it's hard to change those protocols*". Further, this researcher commented that including individuals with IDD in research may change the way they interact with their university's IRB. They discussed IRB regulations and the extensive training required to follow the rules of research, which may be difficult for co-researchers with disabilities to complete:

> Another thing is, IRB board. I think this might change the way I apply for IRB too. Basically, all your concern, you want to ask him, do you want to also be a researcher and then they have to do the training to be a researcher to follow the rules, the

> confidentiality and minimizing the risks. Just getting a little bit complicated situation here. And then they also need..the full faculty members at [university], we kind of have a liability with [university] that we violate, we can get fined. Right. But then, yeah, people outside not with [university]. I think that's another thing we..it's not saying we shouldn't explore. But I think it would create some other things we have to go through to make sure it can happen.

Further, researchers noted that academia tends to be inaccessible for those outside of a university setting, due to its emphasis on advanced literature and intimidating dissemination processes, such as academic papers and presentations. This group noted that while these processes are commonplace for researchers, individuals with disabilities may be hesitant to engage in research if these barriers are in place and may benefit from different forms of dissemination. One researcher noted how conferences can be intimidating for individuals with IDD, and more informal presentations may be beneficial:

> Maybe because of the not so much the attention but just the, the intimidation factor of these are all people that I don't know and they may be professional and I don't feel comfortable talking in front of these people that are presenting this information, but things that are more meaningful for them like presenting information to city council because it directly affects them, their community, their friends and neighbors.

Discussion

Three groups of stakeholders provided their perspective on inclusions of individuals with IDD as co-researchers. In general, all three groups expressed interest in inclusion of those with IDD in the research process. However, all groups identified a variety of barriers noted that make it challenging to conduct community engaged research with those with IDD. Many of these barriers were related to lack of comfort, knowledge and training related to inclusive IDD research. Other barriers were structural and related to the environments in which academic research is conducted.

One commonality between all three groups was an agreement that the inclusion of people with IDD as coresearchers is a worthwhile and important endeavor. For individuals with IDD, research is a platform in which they can express their opinions and contribute to outcomes that matter to them. Participants with IDD felt excited to participate in research where they could talk about their experiences and have an impact on the lives of other individuals with IDD. Similarly, people who support individuals with IDD discussed the inclusion of individuals with IDD as empowering for the individuals they support. When individuals with IDD are invited to advocate for themselves and help shape research, they feel empowered to continue sharing their experiences and engaging with research. Researchers also noted an interest in including individuals with IDD in research, and that it has been useful in the past to include the lived experiences of individuals with IDD when planning projects and interpreting data. Further, multiple researchers expressed that they should be doing more to include individuals with IDD in their research, demonstrating an awareness of the value of lived experience.

While the importance of individuals with IDD as co-researchers was recognized in each group, many discussions turned towards the broad range of barriers that make including individuals with IDD difficult. One such barrier is a lack of comfort related to the inclusion of individuals with IDD. For example, individuals with IDD expressed uncertainty and feelings of intimidation when thinking about participating as a co-researcher. For most individuals with IDD, helping with research is a novel task, and individuals may not know where to start or feel confident enough in their skills to believe that they could provide a meaningful contribution. Similarly, researchers expressed a lack of comfort in including individuals with IDD in their research due to uncertainty about how to effectively include this population in various research tasks. These concerns may demonstrate a need for increased training for both groups to prepare them to effectively participate in inclusive IDD research. This may include developing formal training for self-advocates to better understand what research is, how specific tasks should be conducted and how to contribute to the process. Researchers may also benefit from training on working with those with IDD, how to develop accessible materials, and tailor tasks to the skills of the individuals serving as co-researchers. Additionally, there may be a benefit to promoting more informal meetings between individuals with IDD and researchers. Informal interactions can help build rapport, develop trust and openness, create opportunities to discuss support needs and how to increase comfort with engaged research.

Another notable finding from this study was the potential role of care providers. Individuals with IDD expressed an interest in having trusted individuals, such as family members, staff, mentors or friends, around as they helped with research in the event they needed support or felt uncomfortable. Similarly, individuals from the supports group talked about the importance of creating an open environment that makes individuals with IDD comfortable. This group discussed that when individuals with IDD know they are supported, they are more willing to do things that are out of their normal routine. Support providers also noted that many individuals with IDD have transferable skills that could support research endeavors. This suggests that care providers could be important allies in supporting inclusive IDD research. Given that direct support providers are charged with implementing person-centered plans of care that allow the individual with IDD to engage in desired activities, they may be able to help build engaged research activities into care plans. Other service systems could also be supportive. For instance, Vocational Rehabilitation provides job coaching and other supports for those with IDD to promote competitive employment. These, and other support systems could be particularly important to supporting inclusive research given

their roles. This would necessarily include other natural supports, such as family members or guardians who also support loved ones with IDD given that they may have some degree of formal or informal control over some aspects of the individuals' day-to-day life (e.g., guardianship).

While there was general support for individual measures, such as training or supported employment, that could promote inclusive IDD research, there were also several important structural barriers that could present significant disincentives. Researchers were the group that were most likely to note these challenges, which included a lack of time and resources, limited funding, and institutional limitations such as strict IRB requirements. Structural barriers may be more resistant to change given that they require institutional and/or cultural change. This may discourage researchers, who may have requirements from their institutions to publish papers and acquire grant funding, from attempting CBPR and including individuals with IDD in the research process. This may be especially true for newer researchers who have increased grant and manuscript production demands to secure tenure.

Given the importance of securing funding to advance research, it may be useful for funders to encourage more engaged research, or roles for those with lived experience. While there are some funding mechanisms designed specifically for CBPR, monetary and time costs may still dissuade researchers from pursuing this work if they have institutional demands for research production. Nonetheless, securing significant funding for inclusive and engaged research can help institutions understand the value of this approach and help support researchers that wish to pursue this type of work. Moreover, as more funders recognize the value of engaged research, they may consider building structural features into the award, such as additional time for engagement, CBPR training expectations, and requirements to include co-researchers.

Similarly, the comments related to compliance challenges (e.g., IRB) suggest that research institutions, themselves, may pose significant structural barriers to inclusive IDD research. Certainly, IRBs and research institutions are critical partners in protecting human research subjects and must continue to ensure that research is conducted in ways that do not pose unacceptable risks. However, there may be novel accommodations that IRBs and research offices can make that will allow for more opportunities for co-researchers with IDD, while ensuring the highest level of human subjects protections. For instance, models of co-researcher training that are specifically designed for individuals with IDD while meeting all standard ethical standards [e.g., (27, 28)] can be adopted by IRBs to allow for those with IDD to participate more easily. Trainings such as these contain all necessary learning modules, but do not have the same reliance on inaccessible written language or technical jargon that may create significant barriers to non-professionals. While IRB protocols are vital to protect individuals with disabilities (29), new approaches to training those with IDD may be particularly useful in helping this population understand the research process, their role in engaged research, and help them feel confident in their ability to contribute to the research process.

Limitations and future research

Although we found clear and consistent themes in this study, there are several limitations that should be mentioned. First, these results were developed from a small sample, from a limited geological range. This was intentional given that the experiences of individuals with IDD living in rural areas are underrepresented in research. While this approach allowed for better understanding an often-marginalized group, it also limits generalizability. Future research may seek out the experiences of individuals with IDD in additional geographic areas to further validate these findings. However, it will be important to further study the experiences of those living in rural communities. Given the health disparities for all rural communities, finding innovative solutions to healthcare problems is paramount. Moreover, inclusion of those with IDD in the process may allow for the development of solutions that are more effective for the local challenges facing rural America.

Additionally, the current research identified a number of barriers that make the inclusion of individuals with IDD challenging. For instance, Wyoming is a rural state with a sparse population, making recruitment difficult. Additionally, many individuals with IDD did not have access to email or phone calls, further complicating the recruitment process. This further limits generalizability. Additional work is needed to identify how to address these barriers; however, this research is instructive. Through the process of engaging with the states Developmental Disabilities network (i.e., Wyoming Governor's Council for Developmental Disabilities) we were able to secure a significant amount of interest and participation in this work, which would have been otherwise difficult, if not impossible. Engagement efforts, such as this, may be the most effective approach to developing more robust samples of individuals with IDD. However, researchers will continue to need to make efforts to meet potential participants in the communities that they live and work and may need to employ multiple strategies to be successful.

Further, future research is needed to address the barriers noted in this research. For instance, a recurring theme discussed in this research is that individuals with IDD and researchers do not feel completely comfortable engaging with each other. It will be important to develop training or other supports to help each group be more secure in working with the other. This may include trainings to make research in this area less intimidating for individuals with IDD, or to help researchers know how to better interact with this group. Or other forms of support that can be delivered by direct support providers could be useful. The role of assistive technologies could also be important, as well as addressing structural barriers that may discourage researchers from pursuing engaged IDD research. Regardless, future research related to developing research skills and increasing vocational

efforts in research for individuals with IDD would help encourage more engaged IDD research.

Conclusion

This study demonstrates that, despite strong interest, there are many barriers that make it difficult for those with IDD to participate as co-researchers. These barriers will need to be addressed to facilitate the participation of individuals with disabilities as co-researchers in IDD research. While individuals with IDD, support providers, and IDD researchers all value inclusive IDD research, additional training and modifications to research processes are needed to promote inclusive IDD research. Although addressing some of these barriers will likely be difficult in the near-term including individuals with IDD will strengthen the research process and the products that are developed through inclusive research. Nonetheless, this research is instructive regarding the nature of challenges that stakeholders face when trying to create inclusive IDD research. This will help focus efforts on addressing these barriers to promote greater inclusion.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Institutional Review Board University of Wyoming. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

AD: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Validation, Writing – original draft, Writing – review & editing. TB: Conceptualization, Formal Analysis, Funding acquisition, Investigation, Methodology, Resources, Supervision, Validation, Writing – original draft, Writing – review & editing. CH: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Writing – review & editing. EM: Conceptualization, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Writing – original draft, Writing – review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

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Griffiths Scales of Child Development 3rd Edition: normalization for the Brazilian population

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Introduction: Child development must be carefully evaluated, requiring assessment instruments to assess different areas of development. Griffiths Scales of Child Development 3rd Edition (Griffiths III) is used to assess different areas of development in children. This study normalized Griffiths III for the Brazilian population from 0 to 72 months.
Methods: 445 typically developing children from 0 to 72 months, divided into eight groups (from 0 to 6 months; 7 to 12 months; 13 to 18 months; 19 to 24 months; 25 to 36 months; 37 to 48 months; 49 to 60 months; 61 to 72 months) participated. Their tutors answered the anamnesis protocol. Denver II Developmental Screening Test and Griffiths III were applied. Statistical analysis was performed using the Mann–Whitney Test and Spearman's rank correlation coefficient. Normalization followed the criteria of the original scale.
Results: There was a direct and statistically significant correlation between maternal schooling and socioeconomic status; a direct correlation in the performance between the subscales. The normalization table of Griffiths III with the developmental age of children from 0 to 72 months was elaborated through linear progression, calculated using a specific formula.
Discussion: The data collected for the Brazilian population from 0 to 72 months were normalized, following the guidelines and norms of the original Griffiths III.

KEYWORDS

child development, child, cross-cultural comparison, neurosciences, protocols

1 Introduction

Child development is complex and dynamic and influenced by numerous factors, intrinsic and extrinsic. Monitoring child development and performing early and assertive diagnosis, promoting adequate stimulation and guidance regarding the area or areas that are altered, is crucial for a good prognosis.

Focus, dedication, and continuity in studies related to normative child development are necessary. The use of assessment and diagnostic instruments, combined with the subjective clinical assessment of experienced and active professionals, enables one to carefully verify and understand the different areas of child development in a particular and individual way.

The Brazilian scenario in this regard is discouraging, considering the reduced number of standardized and normalized instruments (1–4). As an instrument for assessing and

diagnosing child development, the Griffiths Scales of Child Development 3rd Edition (Griffiths III) (5) is cited, which was cross-culturally adapted to the Brazilian reality in its entirety (6).

In the international scenario, Griffiths III is widely used to monitor the typical development of children aged from 0 to 72 months, as well as to evaluate and diagnose changes in one or more areas of child development in children with suspect or diagnosed syndromes or with risk factors for changes in development (4, 7–25).

A subsequent step in the process of translation and cross-cultural adaptation of assessment instruments is the verification of psychometric properties, among which we highlight, in this study, the normalization. With normalization, it is possible to compare the performance of a child with others, considering the chronological age (26, 27). Thus, the diagnosis becomes more effective and assertive when the normalization occurs in the child's country of origin, considering the different cultures between countries.

Considering these aspects and the possibility of using Griffiths III for formal and instrumental assessment of the development of the Brazilian child population, with specific standards, this study aimed to normalize the Griffiths III for the Brazilian population aged from 0 to 72 months.

2 Method

The study was approved by the Committee for Ethics in Research, respecting resolution 196/96, which deals with research ethics (CAAE:84323718.3.0000.5417).

The Association for Research in Infant and Child Development (ARICD), which owns the copyright of the Griffiths Scales, authorized the study by signing a contract between the researcher, the association, and Hogrefe, the publisher of the Griffiths Scales.

2.1 Participants

A representative percentage of children from the Brazilian population, recruited from different cities in the State of São Paulo, participated in the study. The percentage was outlined after a statistical study considering sample calculation carried out with data from a pilot study, which had 417 participants.

The sample size calculation considered the highest standard deviation between the Griffiths III subscales (A - Foundations of Learning, B - Language and Communication, C - Eye and Hand Coordination, D – Personal, Social Emotional, E - Gross Motor) and overall performance, respecting a 95% confidence interval, using the GPower 3.1.9.2 program.

Participants were typically developing (TD) children of both sexes, aged between 0 and 72 months, representing the Brazilian population in terms of sex distribution and socioeconomic classification. These data were based on information from the Brazilian Institute of Geography and Statistics (28).

As inclusion criteria, the child needed to present typical development; be chronologically aged between 0 and 72 months; not have moderate to severe sensorineural hearing loss; not have visual impairments that would prevent the performance of the proposed procedures; show a normal result on the Denver-II Developmental Screening Test (29); and have the Free and Informed Consent Form signed by their legal guardians.

Participants were divided into eight groups to perform the sample calculation:

- Group 1 - G1: TD children aged between 0 and 6 months;
- Group 2 - G2: TD children aged between 7 and 12 months;
- Group 3 - G3: TD children aged between 13 and 18 months;
- Group 4 - G4: TD children aged between 19 and 24 months.
- Group 5 - G5: TD children aged between 25 and 36 months;
- Group 6 - G6: TD children aged between 37 and 48 months;
- Group 7 - G7: TD children aged between 49 and 60 months;
- Group 8 - G8: TD children aged between 61 and 72 months.

Considering that child development is influenced by several factors and associated with the fact that this study aims to normalize the instrument for evaluating child development, care and attention to three specific aspects, collected in the anamnesis, were necessary to contribute to the analysis of the results: sex; maternal schooling and socioeconomic status.

As for sex, the sample participants were divided into two groups: female and male.

As for the mother's level of education, an ordinal classification was performed according to the following criteria: Illiteracy (1); Incomplete Elementary School (2); Completed Elementary School (3); Incomplete High School (4); Completed High School (5); Incomplete Higher Education (6); Completed higher education (7); Postgraduate (8).

Regarding socioeconomic status, the following classification was applied, considering the number of minimum wages that the family receives monthly: Classification 1: 0–2 minimum wages; Classification 2: 2–5 minimum wages; Classification 3: above 5 minimum wages.

2.2 Procedures

After the signing of the Acquiescence Form by those responsible for the institutions where the data collection was carried out, contact was established with the tutors responsible for the children in the target age group of the study. The tutors who signed the Free and Informed Consent Form were invited to answer the anamnesis protocol, containing information on conditions of pregnancy, childbirth, neuropsychomotor development data, language, speech, hearing, and perception of the participant.

The Denver-II Developmental Screening Test (DDST-II) (29) was applied as an inclusion criterion in the sample, classifying the participants as typically developing children. Children who had a "Normal" result on the DDST-II were evaluated with Griffiths III (5).

The translated and adapted version of Griffiths III for the Brazilian population (6) was applied to participants aged from 0 to 72 months.

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2.3 Data analysis

For the presentation of the data obtained during the structured interview with the tutors of the children, descriptive statistics were used with absolute and relative frequency values considering a significance level of 5% (30): Mann–Whitney Test and Spearman's rank correlation coefficient.

Normalization followed the criteria of the original Scale, through linear progression from one age group to the next (every two months), using smoothed mean and standard deviation values.

3 Results

Initially, 557 participants were evaluated, of which 112 presented a "Risk" result in the DDST-II, being excluded from the sample, considering the inclusion criteria. The sample consisted of 445 children, aged between 0 and 72 months, who presented a "Normal" result in the DDST-II, met the inclusion criteria, and participated in the normalization of the Griffiths III, in the Brazilian reality.

Sample calculations were performed based on the pilot study (417 participants), using the highest standard deviation value observed in group performance. The results were as follows:

G1 – 47 participants presented a higher standard deviation on subscale A with a sample calculation of 52 participants for the final sample;

G2 – 65 participants presented a higher standard deviation on subscale D with a sample calculation of 70 participants for the final sample;

G3 - 52 participants presented a higher standard deviation on subscale E with a sample calculation of 56 participants for the final sample;

G4 - 33 participants presented a higher standard deviation on subscale D with a sample calculation of 38 participants for the final sample;

G5 - 70 participants presented a higher standard deviation on subscale D with a sample calculation of 70 participants for the final sample;

G6 - 80 participants presented a higher standard deviation on subscale A with a sample calculation of 81 participants for the final sample;

G7 - 49 participants presented a higher standard deviation on subscale A with a sample calculation of 50 participants for the final sample;

G8 - 21 participants presented a higher standard deviation on subscale C with a sample calculation of 28 participants for the final sample

The sample distributed in the eight groups was analyzed in terms of sex, female and male (Table 2), and the following results were obtained: G1 - 46,2% ($n = 24$) were female and 53,8% ($n = 28$) were male; G2 – 48,6% ($n = 34$) were female and 51,4% ($n = 36$) were male; G3 - 48,2% ($n = 27$) were female and were 51,8%.($n = 29$) male; G4 – 47,4% ($n = 18$) were female and 52,6% ($n = 20$) were male. G5 - 54,3% ($n = 38$) were female and 45,7% ($n = 32$) were male. G6 - 45,7% ($n = 37$) were female and 54,3% ($n = 44$) were male. G7 - 42% ($n = 21$) were female and 58% ($n = 29$) were male. G8 – 57,1% ($n = 16$) were female and 42,9% ($n = 12$) were male.

Table 1 presents the data on the mother's level of education, considering absolute frequency.

Regarding the socioeconomic status of the sample, it was distributed considering the socioeconomic reality of the Brazilian population. The socioeconomic status was characterized according to the number of minimum wages that the family receives monthly. Below, the data are presented considering each group of the sample:

G1 - 37 participants had a family income from 0 to 2 minimum wages per month, 13 from 2 to 5, and 2 more than 5 minimum wages;

TABLE 1 Distribution of the sample in terms of the maternal level of education.

MLE / Group	1	2	3	4	5	6	7	8
G1	1	4	11	27	4	5	0	0
G2	2	4	11	41	2	7	3	0
G3	4	3	13	27	5	4	0	0
G4	0	3	4	21	1	9	0	0
G5	0	0	8	9	26	4	17	6
G6	0	1	9	7	30	9	24	1
G7	0	0	7	3	17	6	13	4
G8	0	0	2	1	10	3	11	1
Total	7	15	65	136	95	47	68	12

MLE (maternal level of education); 1 (Illiteracy); 2 (Incomplete Elementary School); 3 (Completed Elementary School); 4 (Incomplete High School); 5 (Completed High School); 6 (Incomplete Higher Education); 7 (Completed Higher Education); 8 (Postgraduate); G1 (typically developing children aged between 0 and 6 months); G2 (typically developing children aged between 7 and 12 months); G3 (typically developing children aged between 13 and 18 months); G4 (typically developing children aged between 19 and 24 months); G5 (typically developing children aged between 25 and 36 months); G6 (typically developing children aged between 37 and 48 months); G7 (typically developing children aged between 49 and 60 months); G8 (typically developing children aged between 61 and 72 months).

TABLE 2 Comparison of boys' and girls' performance on the 5 Griffiths III subscales.

Subscale	Sex	Median	25%	75%	*p*-value
A	M	29.5	2	61	0.796
	F	30.0	2	62	
B	M	33.0	2	62	0.773
	F	34.0	2	62	
C	M	32.5	2	67	0.727
	F	35.0	2	67	
D	M	35.0	3	64	0.733
	F	38.0	3	66	
E	M	36.0	3	63	0.974
	F	37.0	3	62	

A – Foundations of Learning Subscale; B – Language and Communication Subscale; C – Eye and Hand Coordination Subscale; D – Personal, Social Emotional Subscale; E – Gross Motor Subscale; M – male (117 participants); F – female (112 participants); 25% - first quartile; 75% - third quartile.

G2 - 49 participants had a family income from 0 to 2 minimum wages per month, 17 from 2 to 5, and 4 more than 5 minimum wages;
G3 - 40 participants had a family income from 0 to 2 minimum wages per month, 13 from 2 to 5, and 3 more than 5 minimum wages;
G4 - 23 participants had a family income from 0 to 2 minimum wages per month, 10 from 2 to 5, and 5 more than 5 minimum wages;
G5 - 19 participants had a family income from 0 to 2 minimum wages per month, 32 from 2 to 5, and 19 more than 5 minimum wages;
G6 - 27 participants had a family income from 0 to 2 minimum wages per month, 39 from 2 to 5, and 15 more than 5 minimum wages;
G7 - 17 participants had a family income from 0 to 2 minimum wages per month, 23 from 2 to 5, and 10 more than 5 minimum wages;
G8 - 5 participants had a family income from 0 to 2 minimum wages per month, 19 from 2 to 5, and 4 more than 5 minimum wages.

Table 2 describes the median values (50%) as well as the first quartile (25%) and third quartile (75%) of the participant's performance in the 5 subscales, after the application of the Mann–Whitney test, for comparison between sexes, considering the *p*-value statistically significant when lower than 0.05.

Table 3 describes the correlation values and the *p*-value, considered statistically significant when it was lower than 0.05, divided by group and in the total sample, for comparison between maternal education and socioeconomic status.

Table 4 shows the correlation between socioeconomic status and performance on the 5 subscales of Griffiths III for the eight groups. The correlation type and statistical significance were analyzed, considering a *p*-value lower than 0.05.

Table 5 shows the correlation between the participant's performance in the 5 subscales of Griffiths III for the eight groups. The correlation type and statistical significance were analyzed, considering the *p*-value less than 0.05.

Table 6 shows the result of the normalization of Griffiths III with the presentation of the equivalences between the Gross Results of the Subscales, the General results, and the Age of Development in the age group from 0 to 72 months.

TABLE 3 Correlation between maternal level of education and socioeconomic status of the total sample as well as of each group individually.

Group	MLE and SS correlation	*p*-value
G1	0.452	0.001*
G2	0.363	0.002*
G3	0.477	<0.001*
G4	0.535	0.001*
G5	0.697	<0.001*
G6	0.399	<0.001*
G7	0.666	<0.001*
G8	0.489	0.008*
Total	0.559	<0.001*

MLE (maternal level of education level); SS (socioeconomic status); G1 (typically developing children aged between 0 and 6 months); G2 (typically developing children aged between 7 and 12 months); G3 (typically developing children aged between 13 and 18 months); G4 (typically developing children aged between 19 and 24 months); G5 (typically developing children aged between 25 and 36 months); G6 (typically developing children aged between 37 and 48 months); G7 (typically developing children aged between 49 and 60 months); G8 (typically developing children aged between 61 and 72 months).
*Statistically significant *p*-value, less than 0.05.

TABLE 4 Correlation between socioeconomic status and performance on the 5 subscales of the Griffiths III, for each group individually.

Group	Values	SS and A	SS and B	SS and C	SS and D	SS and E
G1	Correlation	−0.058	−0.060	0.042	−0.107	−0.129
	p-value	0.678	0.669	0.768	0.450	0.361
G2	Correlation	−0.028	−0.020	−0.069	−0.024	−0.120
	p-value	0.817	0.867	0.564	0.844	0.322
G3	Correlation	−0.228	−0.304	−0.264	−0.325	−0.129
	p-value	0.090	0.023*	0.049*	0.014*	0.343
G4	Correlation	−0.176	0.008	0.074	−0.044	0.096
	p-value	0.290	0.960	0.660	0.789	0.563
G5	Correlation	−0.115	−0.016	0.000	0.051	−0.021
	p-value	0.342	0.894	1.000	0.676	0.865
G6	Correlation	−0.009	−0.052	−0.036	−0.035	−0.175
	p-value	0.939	0.647	0.751	0.756	0.119
G7	Correlation	−0.093	0.028	−0.157	−0.075	−0.247
	p-value	0.520	0.848	0.277	0.605	0.084
G8	Correlation	0.118	0.056	0.079	0.181	0.129
	p-value	0.550	0.776	0.689	0.356	0.549

Caption: SS (socioeconomic status); G1 (typically developing children aged between 0 and 6 months); G2 (typically developing children aged between 7 and 12 months); G3 (typically developing children aged between 13 and 18 months); G4 (typically developing children aged between 19 and 24 months); G5 (typically developing children aged between 25 and 36 months); G6 (typically developing children aged between 37 and 48 months); G7 (typically developing children aged between 49 and 60 months); G8 (typically developing children aged between 61 and 72 months); A – Foundations of Learning Subscale; B – Language and Communication Subscale; C – Eye and Hand Coordination Subscale; D – Personal, Social Emotional Subscale; E – Gross Motor Subscale.
*Statistically significant *p*-value, less than 0.05.

4 Discussion

Studies aiming to normalize an assessment instrument require a robust methodology that accounts for variables potentially affecting the instrument's application results. Thus, before the application, the instrument is adapted or prepared in a manner consistent with the culture of the population to which it will be applied (2, 31, 32).

Griffiths III was translated and cross-culturally adapted into the Brazilian Portuguese language (6), following a rigorous and careful process, based on the methodology proposed by Beaton (31).

Many countries use the Griffiths scales, such as Germany, South Africa, East Africa, Australia, Belgium, Cambodia, China, Philippines, France, Spain, Greece, Netherlands, Italy, Macedonia, Malaysia, Portugal, Sweden, Switzerland, Russia, and Thailand.

The establishment of the number of participants in the sample respected the sample calculation based on the highest standard deviation verified, based on the performance of the participants in each subscale, and also on the general performance. This calculation was performed for each of the eight groups into which the sample was divided, considering the age groups. The confidence interval respected in the sample calculation was 95%.

TABLE 5 Correlation between the performance in the 5 subscales of Griffiths III, of the total sample, and for each group individually.

Group	Values	A × B	A × C	A × D	A × E	B × C	B × D	B × E	C × D	C × E	D × E
G1	Correl.	0.920	0.917	0.951	0.905	0.913	0.921	0.909	0.943	0.914	0.920
	p-value	<0.001*	<0.001*	<0.001*	<0.001	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G2	Correl.	0.866	0.871	0.839	0.749	0.876	0.867	0.784	0.852	0.747	0.841
	p-value	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G3	Correl.	0.691	0.763	0.634	0.988	0.800	0.762	0.648	0.823	0.811	0.678
	p-value	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G4	Correl.	0.561	0.585	0.641	0.539	0.528	0.663	0.646	0.662	0.639	0.772
	p-value	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G5	Correl.	0.712	0.774	0.744	0.669	0.764	0.795	0.675	0.812	0.687	0.768
	p-value	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G6	Correl.	0.881	0.836	0.739	0.807	0.808	0.774	0.832	0.718	0.795	0.791
	p-value	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*	<0.001*
G7	Correl.	0.741	0.727	0.350	0.674	0.651	0.310	0.657	0.330	0.718	0.313
	p-value	<0.001*	<0.001*	<0.013*	<0.001*	<0.001*	<0.028*	<0.001*	<0.019*	<0.001*	<0.027*
G8	Correl.	0.338	0.318	0.301	0.054	0.262	0.366	0.315	0.233	0.558	0.413
	p-value	<0.079	<0.099	<0.120	<0.803	<0.178	<0.055	<0.134	<0.232	<0.005*	<0.045*

G1 (typically developing children aged between 25 and 36 months); G2 (typically developing children aged between 37 and 48 months); G3 (typically developing children aged between 49 and 60 months); G4 (typically developing children aged between 61 and 72 months); A – Foundations of Learning Subscale; B – Language and Communication Subscale; C – Eye and Hand Coordination Subscale; D – Personal, Social Emotional Subscale; E – Gross Motor Subscale; Correl. (correlation).
*Statistically significant *p*-value, less than 0.05.

Sampling is a highly developed and sophisticated field of statistics that studies the research planning technique. It enables inferences about a universe from the study of part of its components. As sampling seeks the inference from a strictly statistical point of view, the procedure will only be valid if the sample is representative of the population. The sample calculation aims to reduce the costs and time of the estimation process, maintaining a safe minimum precision (33).

Regarding sex, maternal level of education (Table 1), and socioeconomic status, the distributions of the participants were presented. These factors were selected and included in the data analysis, considering the analyses performed in the Griffiths III Manual (5). Studies with GDMS also considered these variables (4, 9, 34–37).

DDST-II is a screening test widely used to screen children aged from 0 to 72 months, with "Normal" or "Risk" results. Due to its wide application and use by our research group, and also for its recognition in the scientific, clinical, and research community, this was the instrument chosen to select the sample (38–40).

Two hundred and fifteen girls and two hundred and thirty boys were evaluated. There was no statistically significant difference in performance between girls and boys (Table 2), analyzing the performance in each Griffiths III subscale individually. There are studies in the literature that corroborate this finding (5, 9, 34).

When analyzing the correlation between the maternal level of education and socioeconomic status (Table 3), there was a direct and statistically significant correlation in the eight individual groups and, therefore, in the total sample. According to the IBGE's Synthesis of Social Indicators (2010), the two correlated variables are directly proportional, which shows the possibility of considering only one of them in further analyses. Thus, socioeconomic status was chosen to analyze its correlation with participants' performance in the Griffiths III subscales.

Table 4 shows the correlation between SS and the performance of the participants in the five subscales of Griffiths III (A, B, C, D, and E), for each group individually (G1, G2, G3, G4, G5, G6, G7 and G8). A statistically significant correlation was observed between SS and the performance in the subscales B, C, and D, only for G3 (from 13 to 18 months). The correlations were weak, some being direct and others inverse. It is noteworthy that only in group 5 (from 25 to 36 months), for SS and Eye and Hand Coordination, there was no correlation. Group 8 (from 61 to 72 months) was the only group that showed a direct correlation between SS and all subscales (A, B, C, D, and E). Groups 2, 3, and 6 (from 7 to 12; 13 to 24; and 37 to 48 months) showed an indirect correlation between SS and all subscales. There was heterogeneity regarding the correlation of SS and performance on the subscales, according to the age group of the participants.

The result of the statistical analysis regarding the correlation between SS and the performance of the participants is different when compared to some studies in the literature (7, 41).

Considering the aim of the study of normalizing an assessment instrument, the sample was not distributed homogeneously in the three levels of socioeconomic classification analyzed (1, 2, and 3), which differs from studies found in the literature that distributed the sample evenly among socioeconomic levels, enabling the analysis of the interference of this factor in the participants' performance.

The correlation between the performance of the participants in the five subscales, among themselves, for the eight age groups, was analyzed (Table 5). A direct correlation was found between all subscales, in the eight groups, as well as in the total sample. In groups 1; 2; 3; 5 and 6 (from 0 to 6; 7 to 12; 13 to 18; 25 to 36; and 37 to 48 months) individually, all correlations were strong, direct, and statistically significant.

There was a direct and statistically significant correlation between all subscales of group 4 (from 19 to 24 months), although this correlation varies between 0.528 and 0.663, in performance comparisons between subscales, not indicating as strong a correlation as in the other analyses.

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TABLE 6 Equivalence between gross results in Griffiths III and Age of development.

Gross result	AD A	AD B	AD C	AD D	AD E
1	<15 days	<15 days	<15 days	<15 days	<15 days
2	1 m	<15 days	<15 days	<15 days	<15 days
3	2 m	1 m	1 m	1 m	1 m
4	3 m	2 m	2 m	2 m	2 m
5	4 m	3 m	3 m	3 m	3 m
6	$4^{1/3}$ m	4 m	$3^{1/2}$ m	4 m	$3^{1/2}$ m
7	$4^{2/3}$ m	$4^{1/2}$ m	4 m	$4^{1/2}$ m	4 m
8	5 m	5 m	$4^{1/2}$ m	5 m	$4^{1/2}$ m
9	$5^{1/3}$ m	$5^{1/2}$ m	5 m	$5^{1/2}$ m	5 m
10	$5^{2/3}$ m	6 m	$5^{1/2}$ m	6 m	$5^{1/2}$ m
11	6 m	7 m	6 m	$6^{1/2}$ m	6 m
12	7 m	8 m	$6^{1/2}$ m	7 m	$6^{1/2}$ m
13	8 m	9 m	7 m	8 m	7 m
14	9 m	$9^{1/2}$ m	8 m	$8^{1/2}$ m	8 m
15	10 m	10 m	9 m	9 m	9 m
16	$10^{1/2}$ m	11 m	10 m	$9^{1/2}$ m	$9^{1/2}$ m
17	11 m	$11^{1/3}$ m	$10^{1/2}$ m	10 m	10 m
18	12 m	$11^{2/3}$ m	11 m	$10^{1/2}$ m	11 m
19	13 m	12 m	$11^{1/2}$ m	11 m	12 m
20	14 m	13 m	12 m	$11^{1/2}$ m	$12^{1/3}$ m
21	15 m	14 m	13 m	12 m	$12^{2/3}$ m
22	16 m	$14^{1/3}$ m	$13^{1/2}$ m	$12^{1/2}$ m	13 m
23	17 m	$14^{2/3}$ m	14 m	13 m	14 m
24	18 m	15 m	15 m	$13^{1/2}$ m	$14^{1/4}$ m
25	19 m	16 m	16 m	14 m	$14^{1/2}$ m
26	20 m	17 m	17 m	15 m	$14^{3/4}$ m
27	21 a 23 m	18 m	18 m	16 a 18 m	15 m
28	24 m	19 e 20 m	19 m	19 m	16 m
29	25/26 m	21 m	20 à 22 m	20 m	17 m
30	27 m	22 m	23 m	21 m	18 m
31	28 m	23 m	24 m	22 m	19 m
32	29/30 m	24 m	25 m	23m	22 m
33	31 m	25 m	25 m	24 m	21 e 22 m
34	32 m	26 m	26 m	25 m	23 m
35	33/34 m	27 m	26 m	25 m	24 m
36	35/36 m	28 m	27/28 m	26 m	25 m
37	37/38 m	29 m	29 m	27/28 m	26 m
38	39 m	30 m	30 m	29 m	27/28 m
39	40 m	31/32 m	31/32 m	30 m	29/30 m
40	41/42 m	33 m	33 m	31 m	31/32 m
41	43 m	34 m	34 m	32 m	33/34 m
42	44 m	35 m	35/36 m	33 m	35/36 m
43	45 m	36 m	37/38 m	34 m	37/38 m
44	46 m	37/38 m	39/40 m	35 m	39/40 m
45	47 m	39 m	41/42 m	36 m	41/42 m
46	48 m	40 m	43 m	37 m	43/44 m
47	49 m	41/42 m	44 m	38 m	45/46 m
48	50 m	43 m	45 m	39/40 m	47 m
49	51 m	44 m	46 m	41/42 m	48 m
50	52 m	45/46 m	47 m	43/44 m	49 m
51	53 m	47/48 m	48 m	45/46 m	50 m
52	54 m	49/50 m	49 m	47/48 m	51 m
53	55/56 m	51/52 m	50 m	49/50 m	52 m
54	57/58 m	53/54 m	51 m	51 m	53/54 m
55	59/60 m	55/56 m	52 m	52 m	55/56 m
56	61 m	57/58 m	53/54 m	53/54 m	57/58 m
57	62 m	59/60 m	55/56 m	55/56 m	59/60 m
58	63 m	61/62 m	57/58 m	57/58 m	61/62 m
59	64 m	63/64 m	59/60 m	59/60 m	63/64 m
60	65/66 m	65/66 m	61/62 m	65/66 m	65/66 m
61	67/68 m	67/68 m	63/64 m	67/68 m	67/68 m
62	69/70 m	69/70 m	65/66 m	69/70 m	69/70 m
63	71/72 m	71/72 m	67/68 m	71/72 m	71/72 m
64			69/70 m		
65			71/72 m		

AD – Age of Development; A – Foundations of Learning Subscale; B – Language and Communication Subscale; C – Eye and Hand Coordination Subscale; D – Personal, Social-Emotional Subscale; E – Gross Motor Subscale.

In group 7 (from 49 to 60 months), strong correlations were observed between subscales A (Foundations of Learning), B (Language and Communication), C (Eye and Hand Coordination), and E (Gross Motor). When subscale D (Personal, Social Emotional) was analyzed with the other four subscales, the correlations were weak, despite being direct and statistically significant.

Correlations in group 8 (from 61 to 72 months) were weak and significant only for subscale C (Eye and Hand Coordination) and (Gross Motor) and for subscale D (Personal, Social-Emotional) and E (Gross Motor). Group 8 showed the lowest standard deviation in the statistical calculation. Thus, it was the one with the lowest number of participants in the sample. This fact may have influenced the result of the correlation, in addition to emphasizing the need for a thorough analysis of which activities/skills were expected in this age group, since it is configured in the last age group of Griffiths III.

The literature describes the influence of the performance of a development area on other areas (34, 42–44). The importance of early diagnosis to assess alteration in one or more areas of child development so that it is possible to provide essential stimulation to the child, preventing negative interference in other areas that are not altered, is unanimous (3, 20, 22, 37, 45–47).

Statistical analysis for data normalization (Table 6) was applied, in which the Child's Age of Development was provided as a result, through the linear progression of an age group for the next (every two months), using smoothed mean and standard deviation values, calculated from a specific formula used in the original Scale (3). The raw data from the normalization process, as well as the child's developmental age (Table 6) will be made available to the statistician responsible for the statistical analysis of the original scale, who will process the data to prepare the other results for the development of the scale in the Brazilian Portuguese version.

In the final normalization of the Griffiths III reference tables, reference values of the Scaled Index, Development Quotient with 95% Confidence Interval, Stanine, and Percentile will be generated, all associated and interconnected.

This process will occur concurrently with the period of translation of the Griffiths III Manuals into Brazilian Portuguese and other bureaucratic procedures so that in a not-distant future

the instrument for the application of the Griffiths III can be made available for use in Brazil.

5 Conclusion

The normalization of the data collected for the Brazilian population aged from 0 to 72 months was performed, following the guidelines and norms of the Griffiths III. Soon, Brazilian health professionals who work with children will have the opportunity to assess child development using this scale, diagnosing global changes in child development or specific areas. The application of the scale and the analysis of the results, appropriately, combined with clinical evaluation by an experienced professional, will provide assertive early diagnoses and essential stimulation, prioritizing the period of greatest brain plasticity in the child.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Bauru School of Dentistry Research Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

AF-V: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. ER: Data curation, Formal Analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. EG: Data curation, Formal Analysis, Investigation, Methodology, Validation, Writing – original draft, Writing – review & editing. DL: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

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

Publisher's note

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Adolescent to adult health care transition for persons with intellectual and developmental disability: current barriers, next steps

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The transition of health care from adolescence to adulthood is a challenging time, especially for persons with intellectual and developmental disability (IDD), where navigating health care transitions can be particularly difficult. Persons with IDD are an especially vulnerable population and they and their caregivers encounter barriers in obtaining high quality health care transition. These barriers result in the suboptimal utilization of health care transition services and consequent poorer health outcomes. Herein, we discuss barriers to obtaining high quality pediatric to adult transitional health care for persons with IDD. We then discuss next steps, some of them well recognized and others underappreciated, for addressing these barriers and thereby achieving an important public health need: the attainment of high quality pediatric to adult health care transition for persons with IDD.

KEYWORDS

intellectual disability, developmental disability, intellectual and developmental disabilities, health care transition, adolescent to adult, pediatric to adult

Introduction

The health care transition from pediatric to adult health care is an important aspect of the provision of good health care. The seminal reports, "Supporting the Health Care Transition from Adolescence to Adulthood in the Medical Home", provide a framework for preparing adolescents for transition, beginning between ages 12–14 and concluding when patients are between ages 18–26 (1, 2). This framework emphasizes a planned process for youth and young adult empowerment, self-determination, and self-management and the achievement of successful integration into the adult health care system.

The process of pediatric to adult health care transition is important for all adolescents and young adults and can be a challenging process for many. Studies on transition outcomes, while of variable quality, indicate that structured transition interventions often result in positive transition-related outcomes; conversely, an absent or unstructured health care transition experience is associated with care gaps, poorer

health and increased rates of hospitalization (3–7). Yet, in the U.S., only about 20% of youth receive necessary health care transition preparation (8).

Persons with intellectual and developmental disabilities (IDD), a population group that is variably defined (9) but whose "membership", however constructed, is characterized by multiple vulnerabilities across the lifespan, including the receipt of good quality health care (10–12). The successful transition to adult health care is especially needed but is often particularly challenging for youth with IDD as they have unique health differences and health care needs relative to their peers in the general population. Intellectual disability, if present, may affect one's ability to fully participate in health-related decision making and navigate the transition process and the adult health care system. Additionally, youth with IDD are more likely to have one or more serious medical, neurological and/or mental health co-occurring conditions (13–17). These factors render young adults with IDD particularly vulnerable to care gaps. Absent or poorly integrated health care transition in some instances may be the cause and in many instances is likely contributory to the development of multimorbidity for adults with IDDs (17–22).

For many reasons, described below, most adolescents and young adults with IDD do not receive formal or adequate transition planning (23–25). In this Perspective essay, we provide an overview of the many barriers to obtaining high quality pediatric to adult health care transition for persons with IDD and then discuss approaches for addressing these barriers.

Challenges in pediatric to adult health care transition services for persons with IDD: key parties and conceptual framework

In discussing the challenges and frank barriers involved in obtaining high quality pediatric to adult health care transition services for persons with IDD, it is useful to begin by considering the diversity of the people accorded this designation. The spectrum of adaptive and intellectual functioning and associated medical and non-medical needs of youth with IDD is extensive (13–17). The designation of IDD encompasses persons with a vast range of phenotypes in the areas of cognition and communication, behavior, sensory deficits and needs, mobility and self-care skills, as well as a large spectrum of medical therapies and requirements for technology supports. Similarly, it includes persons with a vast range of hopes and dreams for their lives. Some of the conditions within the IDD designation are inherited, others acquired; some are present prenatally or around the time of birth and others present later in childhood. Some are associated with static neurodevelopmental courses, whereas others are associated with developmental or neurological regression. The diversity of phenotypes contained within the designation IDD creates significant challenges when developing care strategies for this population. For example, the spectrum of cognitive and communicative abilities, ranging from non-verbal status with profound intellectual disability to persons with mild intellectual impairment and highly fluent communication skills, requires different adaptations to meet the health care transition needs of patients and their families. There similarly is a broad spectrum of behavioral phenotypes as well as physical and medical differences and associated needs. Consequently, any program for the transition of care for persons with IDD from adolescence to adulthood should account for the varied cognitive, adaptive, and medical phenotypes of those with IDD or of the subpopulation of those with IDD served by their program, as well as their non-medical interests and needs, thereby facilitating a person-centered approach to their care.

There are multiple parties central to the health care transition process for persons with IDD. Some of the principals include the family members or guardians of persons with IDD (henceforth, caregivers), primary and specialty clinical care providers, clinical and administrative support staff, allied health professional specialists, care coordinators, social workers, legal professionals, hospital administrators and still others. The perspectives of all of the parties noted above are vital for the design and operation of effective health care transition programs. The incorporation of the diverse perspectives of these different stakeholders not only facilitates a person-centered approach to care for youth and young adults with IDD but also supports important family- and systems-related needs.

There are varied ways to analyze the challenges involved in achieving high quality health care regarding pediatric to adult transition for persons with IDD. The approach used here is to note the challenges when viewed from the perspectives of three groups of principal stakeholders: the adolescents/young adults with IDD, their caregivers, and their clinical care providers. We find considerable overlap in the barriers that they describe. We then reframe these challenges into two broad categories: system of care level barriers and societal level barriers. Finally, we discuss next steps, some of them well recognized and others underappreciated, for addressing these barriers.

Barriers to quality pediatric to adult transition health care: the perspectives of participating youth and young adults with IDD

This level of analysis refers to the personal challenges that individuals with IDD encounter as they transition from pediatric to adult health care services. A large variety of challenges have been voiced. Reported challenges, stated by persons living in different regions and who receive care through varied clinical systems and who have diverse physical and neurodevelopmental phenotypes, include complaints of incomplete understanding of the transition process, low expectations of themselves, perceptions of low expectations by others, feelings of abandonment as they transition from pediatric providers, and a desire for and sometimes conflict in seeking decision making independence from caregivers. They also reported inadequate involvement in the health care planning process, a lack of information about the transition process and related resources,

difficulties forming relationships with new providers, limitations in self-advocacy skills needed to navigate adult medical culture, fatigue from seeing doctors or from seeing too many doctors, inadequate supports in the transition process and for self-management, a lack of coordination of the transition process and in adult health care services, a lack of expertise of some of their health care providers, challenges related to transportation, and challenges with insurance/financial matters (26–35). Racial discrimination has also been voiced as an important impediment in transition health care (36).

It is important to recognize that the barriers that individuals with IDD face while navigating the health care transition process are always superimposed on their daily life experiences. For many this includes daily challenges related to physical or mental health differences, as well as possible challenges related to family dynamics, poverty, discrimination or still other factors.

Barriers to quality pediatric to adult transition health care: caregivers' perspectives

This level of analysis refers to the problems that caregivers face in assisting their children's transfer of care to adult health care services. While many of the reported challenges are also voiced by patients, some are specific to caregivers. The challenges noted below are reported by parents caring for children with diverse forms of IDD from diverse health care systems and countries.

Caregivers voiced numerous difficulties pertaining to their children's health care transition process, including experiencing significant emotional and physical health stresses. Many studies noted that they reported a sense of abandonment by the health care system and feelings of both isolation and of vulnerability of their children (27, 37–39). Some mentioned the burden of additional competing obligations to others in the family or impacts on their own health, work and still other aspects of their lives (37, 38, 40, 41).

The stresses they reported related to the shortcomings of the transition process and of the adult health care system for their children. There were many reports of inadequate provision of information regarding the transition process (27, 39, 42, 43) or that the transition planning took place at too late a time (44). Inadequate information about financial considerations in the transfer of their child's health care to the adult medical system or the expense of needed care for the young adult were also noted as barriers to accessing services (43).

Issues regarding medical decision making for young adults with IDD can be complex and some caregivers also reported not receiving adequate information regarding these issues (44). Related to this, some caregivers reported concerns about relinquishing control over the medical care of their young adult children, worrying that their children may not be able to adequately manage their care. The complexities of transitioning medical decision making to the young adults with IDD was sometimes a source of tension between different dyads of stakeholders (43). For some, the achievement of independent medical decision making for young adults with IDD was not uniformly held to be as high a priority as other objectives of the transition process; this presumably largely related to limited cognitive function of those having severe or profound intellectual disability (45). Some caregivers voiced feelings of frustration associated with a perceived lack of respect for them and their expertise in caring for their children (38, 40, 42, 44).

Most studies indicated parental frustration of the inadequacies and inefficiencies of the services needed for the care of their children. One frequently voiced critique was insufficient coordination of the transition process for the patient and fragmentation of his/her clinical care, sometimes accompanied by poor coordination among adult medical services (37–39, 41, 43–45). This was described as a health care culture shift for their children, from their previous more holistic care centered in a pediatric medical home to a more episodic and reactive mode of adult care. Many caregivers reported needing to serve as a transition coordinator for their children, some even having to battle for services during the health care transition process that were essential for the health of their children (38, 41). Caregivers cited inadequate resources within these systems to meet the complex needs of their children including insufficient equipment and facilities and difficulties in accessing primary care and medical specialists knowledgeable or willing to care for their children with IDD (27, 37, 38, 41); these concerns were amplified in the case of some rare disorders and for families in rural areas that had shortages of providers and programs (43, 44). Caregivers for those with rare disorders associated with IDD noted that the state of incomplete knowledge about the natural history and treatment of many of these conditions adds additional complexity in the care of their children (44).

The critiques of the pediatric to adult health care for their children with IDD extended beyond the medical system. Concerns were voiced about the functioning of the medical, education and community systems as separate silos having minimal collaboration with each other, despite needing to work in an integrated manner to adequately provide for the needs of the young adults with IDD (39, 40, 44). Young adults with IDD were sometimes "left floundering" in adulthood for considerable periods of time, waiting for receipt of adult services or other community supports (27, 37–40, 42–44).

Analogous to the health care transition experiences of their loved ones with IDD, the challenges experienced by caregivers can be exacerbated (or mitigated) by other family, community, and societal factors. Some caregivers specifically noted the value of informal support networks or formal parent support or disease-specific support groups in providing emotional support and/or useful information about the transition experience (37, 40).

Barriers to quality pediatric to adult transition health care: clinical care providers' perspectives

Clinical care providers in the pediatric to adult health care transition process are a diverse group of clinicians and include

primary care and various clinical specialty providers. Like the other major stakeholders in this process, providers report multiple challenges in accomplishing effective transition services for persons with IDD. Some providers noted insufficient knowledge of both the transition process and of the unique needs of persons with IDD that are required to adequately engage their clients in the transition process. Salient examples include insufficient knowledge or comfort with clinical management or guardianship-related matters (32, 46). These limitations can be particularly pronounced in the clinical management of youth or young adults with IDD with medical and/or mental health complexity.

Clinical providers also reported that there are insufficient numbers of both primary and specialty health care providers with training or expertise in treating patients with IDD and associated conditions (34, 46–48). This problem of insufficient availability of appropriate clinicians to serve IDD youth can be exacerbated in rural and other medically underserved areas. An additional state or country-specific concern is the difficulty in accessing appropriate generalists or subspecialists because of various health insurance barriers. Many persons with IDD in the United States, for example, are served by public insurance and this, in turn, limits access to many clinical care providers, especially for adults with IDD.

Persons with IDD may have challenges in understanding complex information and in expressing themselves and may also have complex medical histories. These issues necessitate increased time for medical encounters. In usual medical practice, however, clinicians report that they typically are not provided additional time during clinic sessions for this patient population (39, 46, 48). Moreover, while optimal clinical care for medically complex patients requires consultation and coordination with multiple medical services, some medical reimbursement systems, such as in the United States, have hurdles in enabling financial coverage for meetings involving multiple clinicians (39, 46, 48). This issue extends to other needed non-medical consultations that are important in enabling effective transition programs, such as social work and legal consultations for the affected individuals and their family members. The substantial financial debt incurred by some clinicians during their training can also impact their willingness to be devoted to caring for patients with IDD whose care may be less remunerative and associated with other perceived challenges (48).

Providers also noted various educational and administrative barriers that are imposed on their clinical practices. For some this included insufficient or suboptimal educational materials for themselves or their patients, difficulty with electronic medical record systems in including relevant information, as well as difficulties in communicating with clinical care providers working in different medical systems (32, 39, 46, 49, 50). Some noted a need for standardized surveillance and management guidelines in their fields (47, 48). An inadequacy of relevant training for medical students and training programs for residents, fellows, and established practitioners in many clinical fields was also reported (47–49). Providers also reported insufficient administrative and financial support for pediatric to adult transitional health care efforts by the hospitals or clinics that they practice in or by their governments (47, 50).

Some clinical care providers cited family- and patient-related barriers to achieving effective transition. Examples included difficulties or resistance to transition by patients or caregivers because of strong bonds with the pediatric service or fear of leaving the pediatric system; economic issues of families and patient crises were other factors that could be barriers to effective clinical transition (46, 48, 50).

System of care and societal level challenges to obtaining quality pediatric to adult transition health care

The barriers to obtaining quality pediatric to adult health care transition experienced by patients, their caregivers and their clinical care providers can, with only a few exceptions, be largely categorized into two broad areas: barriers at the system of care level and societal level barriers. System of care level barriers refer to barriers that are inherent within various systems of care that impede the transfer of pediatric to adult health care for persons with IDD. Pertinent systems of care include health care systems and aspects of educational and legal systems. These systems of care exist in the context of local, regional, and national governmental resources and regulations that are specific to the location of the affected individual and their family. Depending on the patient's location, therefore, there may be greater or lesser access to associated important resources, thereby impacting both medical and non-medical dimensions of life of the individual with IDD.

Societal level barriers refer to barriers that are a "higher" level of barrier. They are pervasive throughout systems of care and are embedded within the social, economic and political structures of a society. Important societal level barriers include poverty, income inequality, various forms of systemic discrimination and bias and geography-related barriers.

Numerous and varied system of care level barriers were voiced by persons with IDD, caregivers, and clinical care providers. These are noted in the previous sections of this essay and listed in Supplementary Table S1, along with their known or proposed causes and proposed interventions for their amelioration. In general, the system of care barriers include limited availability of structured pediatric to adult health care transition programs, problems with access to care for persons with IDD due to health care provider shortages or inadequate expertise of clinical providers, inadequate time for the clinical encounters, inadequate information and other resources to support the transfer of care for persons with IDD and their families, inadequate coordination of information and of clinical care within and between systems of care, inequitable access to office care related to scheduling or the physical design of clinics or transportation to clinics, financial challenges relating to the coverage of clinical care and related services that impact patients and caregivers, and financial challenges that relate to professional remuneration and that impact clinical providers and their administrators and clinics/hospital system.

There also are diverse, important societal barriers to pediatric to adult health care transition, including but not restricted to those with IDD. These are especially apparent through formal epidemiologic analyses and are also listed in Supplementary Table S1. Socioeconomic differences, the language used in the household, sex, and race/ethnicity impact health care transition disparities for youth (8). Low-income adolescents in the U.S. with IDD have been reported to receive less health care transition services compared to higher income peers (24, 25). Minorities, particularly Black and Hispanic patients, are also less likely to receive health care transition services, compared to white peers. Black individuals with IDD cite other systemic inequities, including inadequate diagnostic evaluations and biases within schools and other public programs, as further barriers to care (25, 36, 51). Provider bias and discrimination based on a diagnosis (or perception of the diagnosis) such as a mental health diagnosis in persons with IDD has been noted as well (34). Related studies indicate that the receipt of health care transition services varies substantially for youth with different chronic conditions such as intellectual disability, autism or cerebral palsy relative to persons with other special health care needs (23, 24). Disparities in care that relate to geography, such as particular difficulties regarding access to needed medical care for persons living in rural areas, have been reported. Patients having rare conditions who need access to specialty care that has limited availability and which is often only present in medically resource-rich areas, patients living in remote areas, and patients lacking access to appropriate transportation are particularly vulnerable to geography-related health care disparities (43, 44).

An alternative, additional perspective by which to consider many of the barriers experienced in the health care transition process is that they are consequences of ableism (52, 53). This conceptualization views many of these challenges consequent to past and present policies, institutions and norms that devalue and disadvantage persons who are disabled. It can therefore account, in part, for barriers ranging from inadequate resources for all stakeholders in the transition process, inadequate training for health professionals around working with disabled persons, transportation-related challenges for patients and caregivers, difficulties with communication access, limitations regarding the accessibility of scheduling platforms, limitations of the physical accessibility of facilities and more (52–54).

Moving forward

These system level and societal level barriers to pediatric to adult health care transition, often leave youth and young adults with IDD unserved or underserved and they and their caregivers unduly burdened with orchestrating a complex transition process. In addition to detailed resources regarding the principles and practical operation of health care transition programs (2, 55), there are excellent, user-friendly resources for health care transition for youth with disabilities, including IDD, that are separately targeted for youth/young adults, parents/caregivers and clinicians/direct service providers (56). Moreover, multiple parties have issued thoughtful recommendations regarding how to remedy the barriers in health care transition experienced by persons with IDD or complex health conditions (2, 5, 44, 57–64). Previously reported or proposed initiatives to remedy these barriers, as well as some of our own, are noted in Supplementary Table S1. Rather than repeat those recommendations that are excellent and generally agreed upon, we briefly discuss several perspectives on solving transition-related barriers that, in our view, are inadequately emphasized in the literature. We then briefly note some underemphasized opportunities to improve aspects of the health care transition process for those with IDD.

Moving forward: some important but underemphasized perspectives

First, on creating or implementing a change in a health care transition program, the insights and needs of all key stakeholders of the transition program ecosystem should be recognized and met for a highly functional, sustainable program to be achieved. To accomplish this, it is essential that all key stakeholders in health care transition programs be meaningfully involved in decision-making processes. This should include persons with IDD to the degree that they are able to participate. The process of involving persons with IDD in the medical decision making process can be complex, depending on the presence and extent of intellectual disability and/or communication impairment and the abilities of caregivers and clinical providers to facilitate the maximum involvement of each patient. The transition program needs to be involved in this process (2, 65).

With respect to the caregivers, their insights about the transition process and their health and wellbeing are also essential. They have special insights about the medical and non-medical needs of their loved ones and participate in their care and transportation; consequently, their input is essential. And their health and other aspects of their lives are intertwined with their children's lives and health care transition; some quality assessments of the transition process include caregiver wellbeing among their parameters (7).

Clinical care providers on both the pediatric and adult sides of the transition process interface with the patients and their families, social workers and hospital administrators and the insights of these providers are therefore invaluable. Clinicians need adequate time to provide good quality clinical care, good communication systems with patients, caregivers and colleagues, adequate clinical space, appropriate clinical equipment and administrative supports.

Hospital administrators make key decisions regarding personnel, allocation of resources and space issues affecting transition programs. Consequently, hospital administrators need to be informed of the goals, needs and progress of the transition programs and, in turn, need to provide feedback from their perspective.

Providing a forum for all stakeholders to meet and collaborate is essential to the success of a transition program. Different agendas and different types and frequencies of meetings are needed to achieve the needs of all stakeholders. Some committees/agendas

speak to the needs of a specific patient/family, others relate to the "global" design and function of the transition program. The particulars of the committees and the frequencies of their meetings will necessarily be context dependent; one size will not fit all transition programs. Yet, regardless of whether the agenda pertains to a specific individual or to the general functioning of the program, there must be meaningful representation of all major stakeholders in decision making processes. The perspective noted here is different than the frequently cited need for "patient-centered" care. We endorse patient-centered care but all major stakeholder needs should be met for a transition program "ecosystem" to be effective and stable over time.

Second, there is still another important layer of complexity in the attempt to have good representation and communication of the major stakeholders in the health care transition process. The foremost priority of a health care transition program is to ensure effective pediatric to adult health care transition. However, all stakeholders recognize that a healthy, comprehensive transition from the pediatric to adult years is not limited to the patient's medical needs but relates as well to the education, vocational or day program, and social/recreational needs of the patient. This, in turn, necessitates at least minimal integration of the special education system, community organizations and local government with the health care transition process. The specifics of such integration will depend on the needs of each patient and the resources of each of the parties and the region/country where this takes place. Nevertheless, it is extremely important in the life of the patient.

Third, a review of the causes of the diverse barriers to health care transition listed in Supplementary Table S1 indicates that a disproportionate fraction of the barriers is, in our view, attributable to insufficient financial resources and other supports. The basis for this is multifactorial and specific to each region/country but often results from the low reimbursement of many clinical and administrative services of transition programs. Regardless of whether every phase of a transition program is well designed, it must have adequate financial and other resources and those resources need to be stably available for that program to be effective in the long term. The development of adequate and stable financial backing for transition programs is complex and requires commitment by hospital administrators, public health policy makers and legislators. It is a fundamentally important issue that is discussed in only a minority of the documents on interventions to improve transition programs—with some very important exceptions (e.g., 2)—but impacts nearly all such programs, albeit less so in medically well-resourced areas served by universal health care systems. Insightful strategies to improve the financial wellbeing of U.S. health care transition programs have been proposed (2).

A fourth high level perspective on issues pertaining to barriers in health care transition is the analysis of many such barriers as being consequent to ableism, as mentioned above. Viewing transition-related barriers in this light also suggests multiple strategies for intervention; some of these involve regulatory and legislative remedies that would likely be location specific (Supplementary Table S1) (52, 54).

Still other actionable transition-related barriers are relatable to ableism. The literature on remedying barriers to health care transition for those with IDD speaks to inadequate numbers of clinicians who are knowledgeable in caring for those with IDD and the need for improved medical education in this regard. While this is true, this gap in health professional education is part of the larger problem of the inadequacy of health professional education regarding disability-related matters more generally (66, 67). This is partly attributable to ableist perspectives in health professional education. Learning in these areas will expand the pool of clinicians competent in caring for those with IDD (and other conditions) and engender deeper understandings of the lives of those with disability for all clinicians.

The literature on remedying barriers to health care transition provides only minimal discussion of issues of transportation to and the physical space of clinical programs. These are issues that relate to the needs of both the patients and families as well as the needs of the clinical staff and are consequent to, at least in part, ableism. In terms of the former, there should be attention to accessibility of the clinic by public transportation and increased use of telehealth, when appropriate. Accessibility of the clinic rooms and other spaces for persons with mobility or other relevant physical limitations is essential. There should also be modifications to accommodate those with special sensory limitations or sensitivities. The clinic space should be equipped with accessible scales, accessible examining tables and other equipment needed to allow for safe and comprehensive examinations of patients; there are recent federal regulations in the U.S. regarding this matter (68).

Moving forward: some underemphasized or relatively new opportunities to improve transition-related barriers

It is important that health care transition programs, like any health care initiative, have quality assurance/quality improvement processes and therein are other opportunities for program improvement. Quality assurance/quality improvement activities serve two critical purposes: to improve individual care and to refine the transition model. These matters are discussed in the literature on measuring outcomes of health care transition efforts (3, 7, 69) but generally do not receive as much attention in the related literature that specifically addresses remedying transition-related barriers. These activities should cover all key aspects of the transition effort, including clinic operations and clinical effectiveness. Examples include wait times, timeliness of transfer, gaps in care such as missed appointments, preventative and medication care management, insurance continuity, cost effectiveness, and longitudinal outcomes and should involve feedback from all parties, especially that of persons with IDD and other key stakeholders. The standardized inclusion of quality assurance/quality improvement processes in pediatric to adult health care transition programs would likely result in multiple significant benefits for patients and the overall transition program.

Moreover and as noted above, relatively recent data indicate disparities based on race, ethnicity, socioeconomic status and

diagnosis regarding the transition from pediatric to adult health care for persons with IDD (8, 24, 25, 51). Consequently, another vital part of a transition program's quality assessment/quality improvement process should be to assess its effectiveness in achieving equitable health care access and outcomes for all adolescents and young adults with IDD, as has been proposed recently (69–71).

Another set of potential opportunities for transition program improvement involves the consideration of the team that works with the patient and his/her caregivers. There are varying designs of successful health care transition programs including, for example, "general" transition clinics, organ system- or disorder-specific transition clinics, and different permutations of consultation transition clinics or transition teams. Staffing, therefore, will necessarily be contingent on the needs of the clinic population, the design of the transition program, and the local availability of personnel. This should include representation from primary care and major specialties pertinent to the patient, as well as community health or social workers who are able to address and provide resources for the medical issues and social factors affecting patients' well-being.

Among specialist care, it is imperative for patients to receive adequate mental health support, especially since mental health often declines during transition (34, 72, 73). The involvement of mental health professionals as central members of a transition program is an important consideration.

The dental needs of all persons with IDD must be met and ideally should be considered within the context of a transition program. As with medical transitional care, there are multiple barriers in obtaining dental transitional care. These include a shortage of general dentists who are willing and able to provide dental care for those with special health care needs and reimbursement-related barriers (74–76). Patients' dental care during transition should not be considered apart from their other medical needs.

Medical geneticists are not commonly included as part of the core health care transition team for those with IDD. This is sometimes because medical geneticists, if part of transition teams, are mainly used in consultative roles in genetic disorder-specific transition programs. In some instances, this may be due to unavailability of such clinicians. Yet these specialists, when available, can serve multiple useful functions in transition programs. A considerable percentage of those with IDD have an underlying genetic etiology for their condition (77, 78). Medical geneticists can assist in the diagnostic evaluation of the many persons with IDD who do not have an etiologic diagnosis. A genetic evaluation can sometimes reveal diagnoses that in their absence would deprive a patient of a useful treatment, such as in some persons mistakenly diagnosed with cerebral palsy (78). Moreover, persons with uncommon or rare cytogenomic or monogenic conditions with IDD can benefit from the inclusion of medical geneticists on the care team because medical geneticists have familiarity with the natural histories and management of these rare conditions. There are means to incorporate the input of medical geneticists into the primary care of the patient even if they are unable to be physically present (79). Genetic counselors can similarly have helpful roles in health care transition programs (80).

Another potential opportunity relates to the use of short videos. Patients, caregivers and clinical providers all reported on difficulties in the communication of clinical information about patients to clinical providers in the adult medical system. An approach that can augment existing methods and that, to our knowledge, is largely unused for this purpose is the use of short videos that contain pertinent medical and non-medical information about the patient. One possible scenario could be the production of two short, inexpensive videos. One of the videos would feature the patient and describe the patient's interests, likes and dislikes, hopes for the future and other general matters and that would be communicated by the patient, if possible, and/or his caregivers. We previously described the rationale and production of a similar video for adults with IDD who are about to live in out-of-home placements (81). A second short video could be made by a pediatric primary care clinician that would contain the summary of essential medical information needed for the adult care team that will be assuming care of the young adult with IDD. This film could include the patient and family as well, if desired.

Another, non-mutually exclusive method of ensuring understanding and communication of a patient's past and current medical condition is the utilization of a "warm handoff" between providers (32). This conversation can be done via telehealth, phone or even in person in a shared clinic visit with both providers, if circumstances allow. This is a method that we have employed, and our patients and caregivers have expressed their appreciation and reassurance following these interactions.

Finally, there are many unanswered questions regarding how best to accomplish effective pediatric to adult health care transitions for persons with IDD and their families. Some important issues include: What are the best health care transition models for persons with IDD and which models are best in consideration of different contexts (e.g., different health care systems, medical resource-rich vs. resource-poor areas, differing IDD clinical phenotypes)? What are the best approaches for integrating health care across community and government health care resources? How can the vocational, social and recreational needs of youth with IDD best be integrated into their pediatric to adult health care transition? What are the best ways to train the next generation of health care providers to become effective clinical practitioners in their areas of health care transition medicine? How can various communication technologies be used to enhance learning and empower persons with IDD to optimize their care? What are the ways that one can measure the different aspects of the health care transition process, including outcomes and quality assurance/quality improvement processes, and which measures are most useful? What are the best systems of region and country-specific government supports to assure effective, sustainable programs? These and many other relevant issues should be addressed through well-designed research studies, many of which have been nicely framed by others (69, 82). Because of the importance of these issues, health care transition programs should endeavor to have a research component to their

operation. This, in turn, will likely fuel new and possibly unexpected advances in the care of this vulnerable population.

Data availability statement

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

Author contributions

SR: Writing – original draft, Writing – review & editing. SS: Writing – original draft, Writing – review & editing. JC: Writing – review & editing. CC: Writing – review & editing. TD: Writing – review & editing. MN: Writing – original draft, Writing – review & editing.

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Conflict of interest

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

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2025.1486325/full#supplementary-material

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The level and factors associated with caregiver experience among parents of children with cerebral palsy: a cross-sectional study in Southwest China

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Background: Disability is a global public health issue, affecting one in seven people worldwide. In China, there are about 5 million children under 14 years of age with cerebral palsy and about 40,000 new individuals occur every year. Previous studies have revealed that the caregivers of children with cerebral palsy were more likely to perceive a greater burden compared with caregivers of typically developing children. However, there is a lack of information available on the care burden experienced by parents of children with cerebral palsy in China.

Objectives: This study was conducted to determine the level of care burden and its related factors in the parents of children with cerebral palsy.

Methods: This descriptive cross-sectional study was conducted with parents of 165 children with cerebral palsy who were enrolled in children rehabilitation departments of three tertiary hospitals between September 2021 and December 2022. Besides demographic information, the Caregiver Burden Inventory, the Patient Health Questionaire-9, and the Fatigue Severity Scale were used to collect data. Data were analyzed by descriptive and inferential statistics (correlation and multiple linear regression analysis).

Results: The mean (±SD) Caregiver Burden Inventory score of the parents was 42.18 ± 18.79. The score of Fatigue Severity Scale and Patient Health Questionaire-9 demonstrated positive moderate to strong correlations with caregiver burden ($r = 0.461$, $p < 0.001$; $r = 0.630$, $p < 0.001$, respectively). The results of the multiple linear regression analysis showed that a low level of education, long caring time combined with visual impairment, higher depression, and fatigue had an influence on caregiver burden, and 46.4% of the variance in caregiver burden was explained by these factors.

Discussion: The key predictors of caregiver burden include the level of education, caring time, children with visual impairment, and the degree of depression and fatigue. Efforts should be made to relieve the burden on parents of children with cerebral palsy.

KEYWORDS

cerebral palsy, child, caregiver experience, parents, depression, fatigue

1 Introduction

Disability is a global public health issue, affecting one in seven people worldwide (1). The World Health Organization reported that about 15% of the world's population was living with disability in the year 2020, including about 93 million children and 720 million adults with significant difficulties in functioning (2). Developmental disorders in children encompass a range of conditions that affect a child's physical, cognitive, or emotional growth and development (3). These disorders can impact various areas, including learning, communication, social skills, and daily functioning (3). Some common developmental disorders in children include Autism Spectrum Disorder (ASD), Down Syndrome, Attention-Deficit/Hyperactivity Disorder (ADHD), Dyslexia, Cerebral Palsy (CP), and so on (4–6). The World Health Organization (WHO) defines "Quality of Life" (QoL) as the individual's perception of their position in life in the context of the culture and value systems in which they live and about their goals, expectations, standards, and concerns (7). Previous studies showed that caregivers of children with ASD and ADHD have a higher score of stigmas, burden, depression, anxiety, and a poorer QoL than normal (8, 9). Surveys revealed that the primary themes of stressful experiences among parents of young children with Down syndrome included emotional burdens, caregiving responsibilities, struggles against stigma and discrimination, concerns about the future, and challenges related to health, education, and financial issues (10–13). Approximately 95% of mothers with dyslexic children experience anxiety about their child's future and academic performance, in addition to distress related to dyslexia (14–16). In China, among the mothers of 252 children with ASD, 26.02, 17.34, and 3.57% had mild, moderate, and severe depression, respectively (17). Caregivers of children with ASD scored significantly lower on adaptability than that of the norm (18), 46% of parents scored above the cut-off for stress (19), the total score of the Zarit caregiver burden Scale for caregivers of children with ASD was reported (52.62 ± 14.65) in Henan Province, China (20). A qualitative study conducted in China revealed that parents of children with ADHD use individual, family, and school coping strategies to cope with their stress (21). A meta-synthesis included eight qualitative types of research indicated that feeding pressures, educational concerns, language difficulties, and discrimination and stigmatization led to psychological, economic, and family stress in caregivers of children with Down syndrome (22). Four key categories of potential factors contributing to parental stress were identified: cultural factors, parents' psychopathological symptoms, problem behaviors in children with developmental conditions, and caregiver burden (23–25).

Cerebral Palsy, with a rate of about 2.5 per 1,000 live births (26, 27). In China, there are about 5 million children under 14 years of age with cerebral palsy and about 40,000 new individuals occur every year (27, 28). Cerebral Palsy is a chronic illness in childhood, defined as a group of functional limitations that stem from challenges in the development of the central nervous systems, the continuous rehabilitation and lifelong support care are essential components of the development plan for a child with cerebral palsy (29).

Parenting a child with CP can be a unique journey filled with both challenges and rewards (30, 31). The key factors that contribute to positive experiences, include support networks, resilience, and personal growth, alongside those that lead to negative experiences, such as emotional and psychological stress, financial strain, isolation, and social withdrawal, navigating healthcare systems, concerns about the future, guilt and balancing family needs, fatigue and burnout, limited personal time, impact on family dynamics (32–35). The high medical expenses and education costs may turn into an economic burden for parents of children with cerebral palsy (28). The continued provision of excessive care leaves caregiver experiencing fatigue and depression, which can negatively impact their quality of life (28, 36, 37). Families may find it challenging to participate in social activities or events due to their child's needs, leading to feelings of isolation or exclusion (38). While there are significant challenges, many parents also discover immense joy, growth, and fulfillment in their journey (39). The key often lies in seeking support, celebrating progress, and finding ways to cope with the difficulties that arise (40). Many parents report a profound emotional connection with their child with CP. The challenges they face together can strengthen their bond and create a deeper understanding of one another (41). Additionally, family members often become more compassionate and supportive, learning valuable life lessons about diversity and inclusion (42). Furthermore, this experience can lead to personal growth and a new perspective on life, prompting parents to develop patience, empathy, and problem-solving skills (43).

Caregiver burden refers to a multidimensional response to physical, psychological, emotional, social, and financial stressors associated with caring experience' (44). Surveys conducted in Brazil (45), Portugal (46), and Turkey (36) have revealed that the caregivers of children with cerebral palsy were more likely to perceive a greater burden compared with caregivers of typically developing children. Within the context of the stress process model, numerous factors may affect the degree of burden for parents of children with cerebral palsy. Stigma is an important source of psychological stress for families of children with disability (47). Regarding caregiver characteristics, several studies show that having a lower income and education, living in a rural area, greater age, and poor health status are related to a higher burden in caregivers (36, 44, 48, 49). In terms of child characteristics, age, number of functional deficits, type of cerebral palsy, degree of disability, Gross Motor Function Classification System (GMFCS) level, and disease severity were associated with higher burden in caregivers (44, 48–51). The caregiver's social support, self-efficacy, anxiety, depression, and fatigue were found to have significant correlations with caregiver burden (36, 45, 46, 48, 50, 52). A recent study reported the COVID-19 pandemic worsened the psychological status and the care burden of caregivers of children with cerebral palsy (53).

In pediatric psychology, examining the role of families can create an opportunity for understanding and treatment of chronic illness is well-documented (46). Bella et al. study has shown the level of burden and psychological maladjustment of parents has been linked to behavioral and emotional disturbance of children (45). Furthermore, screening parents for excessive burden and assisting them in finding appropriate coping mechanisms significantly contributed to the successful management of stressors and better social function in their children (42, 50, 54). In China, however, there is a lack of information available on the care burden experienced by parents of children with

Abbreviations: CP, Cerebral Palsy; GMFCS, Gross Motor Function Classification System; CBI, Caregiver Burden Inventory; PHQ-9, Patient Health Questionaire-9; FSS, Fatigue Severity Scale.

cerebral palsy. Therefore, the main aim of the current study was to determine the levels, correlates, and predictive factors of care burden among parents of children with cerebral palsy in China.

1.1 Research questions

Q1. What is the level of caregiver burden among parents of children with cerebral palsy in southwest China?

Q2. What demographic and socio-economic factors are associated with caregiver burden among these parents? a. How do parental age, education level, and employment status correlate with caregiver burden? b. Does the severity of the child's condition correlate with the level of caregiver burden?

Q3. What depression and fatigue are associated with caregiver burden?

1.2 Hypotheses

H1: The level of caregiver burden among parents of children with cerebral palsy in southwest China is significantly higher than the general population, indicating a substantial impact on their daily lives.

H2: There is a significant positive correlation between the severity of the child's cerebral palsy and the level of caregiver burden, such that parents of children with more severe disabilities report higher levels of burden.

H3: Lower levels of parental education and employment status are associated with higher caregiver burden, suggesting that socioeconomic factors play a crucial role in the experience of caregiving.

H4: Higher levels of depression and fatigue among parents are significantly associated with higher levels of caregiver burden.

2 Methods

2.1 Design

This cross-sectional study was performed in children's rehabilitation departments of three tertiary hospitals in XX between September 2021 and December 2022. A local hospital's Institutional Review Board approved this study.

2.2 Participants

This study was conducted among parents of children with cerebral palsy who were enrolled in children's rehabilitation departments of three tertiary hospitals in Chengdu, Sichuan Province between September 2021 and December 2022. It was aimed that all parents who have a child with cerebral palsy would agree to participate in this study, without using sample selection. A total of 173 parents agreed to participate in the study, 8 parents were excluded because they provided insufficient/unreliable data, and 165 parents were finally included in the data analysis. The inclusion criteria of parents were as follows: (i) Caring for child aged 1–14 with cerebral palsy, (ii) parents who are taking on the responsibilities for the daily provision of care, (iii) living with the child, could read and write in Chinese. The inclusion criteria of children were as follows: (i) the children with cerebral palsy were diagnosed according to the diagnostic criteria of China Cerebral Palsy Rehabilitation Guidelines (2015), (ii) the children were aged 0–14 years old.

All participants self-completed the questionnaire in the presence of the principal investigator. The overall time taken to collect data by completing the questionnaires was on average 30–45 min.

2.3 Measures

Besides demographic information, the Caregiver Burden Inventory (CBI), the Patient Health Questionaire-9 (PHQ-9), and the Fatigue Severity Scale (FSS) were used to collect data. The selection of the CBI, PHQ-9, and FSS is pivotal for gaining insights into the experiences of caregivers, particularly parents of children with cerebral palsy. Each of these tools has been validated and widely used in research, providing crucial data to understand the intricacies of caregiver experiences. By employing these validated instruments, researchers and healthcare providers can identify specific needs and challenges, ultimately leading to improved support systems that enhance the well-being of both caregivers and their children. This comprehensive approach fosters a deeper understanding of the factors influencing caregiver burden, paving the way for effective interventions.

2.3.1 Demographic characteristics of participants

The demographic characteristics questionnaire was composed of two parts. The first part included the basic information of parents, including the relationship to the child (father and mother), age, education level (primary school and below, junior high school, senior high school, university, master and above), occupation, marital status, average caring time (hours/day) and family systems (composed of nuclear family, stem family, joint family, single parent family). The definition of a nuclear family is a family unit that includes two married parents of opposite genders and their biological or adopted children living in the same residence (55). The stem family is composed of parents, unmarried children, and married sons and their wives (56). The joint family consists of parents and their unmarried children, two or more married sons, and their wives (56). The single-parent family consists of at least one dependent child and the mother or father, the other parent being dead or permanently absent (57).

The second part included the basic information about children with cerebral palsy, including age, gender, types of cerebral palsy, Gross Motor Function Classification System (GMFCS), visual impairment, and epilepsy. The types of cerebral palsy are comprised of Spastic hemiplegia, Spastic diplegia, Spastic tetraplegia, Ataxia, Dyskinetic, and Mixed types, according to China Cerebral Palsy Rehabilitation Guidelines (2015) (27). The gross motor skills (e.g., sitting and walking) of children and young people with cerebral palsy

can be categorized into five different levels using a tool called the Gross Motor Function Classification System (58, 59).

2.3.2 Caregiver Burden Inventory (CBI)

The Caregiver Burden Inventory (CBI) is a self-administered, multidimensional scale for assessing caregiver burden among caregivers of persons with dementia (60), children with cancer (61), psychiatric patients (62), and rheumatoid arthritis patients (63). This measure consists of 24 items divided into five domains: Time-dependence (1–5 items), developmental burden (6–10 items), physical burden (11–14 items), social burden (15–18 items), and emotional burden (19–24 items) (64). Each item is rated on a 5-point (0–4) Likert-type scale, with the total score ranging from 0 to 96 (65). The internal consistency of the *Chinese*-CBI was reported as 0.95 in China (64). In the present study, the Cronbach's alpha of the CBI was 0.944, suggesting the internal consistency of the CBI was adequate. The CBI has been validated in various populations, showing strong correlations with other measures of psychological distress and quality of life (66). This established correlation makes it a reliable instrument for assessing caregiver experiences, particularly in parents of children with CP.

2.3.3 Patient Health Questionaire-9 (PHQ-9)

The Patient Health Questionaire-9 (PHQ-9) is composed of nine items. It can be used to assess depressive symptoms of the general population over the last 2 weeks (67). The total score ranges from 0 to 27; a score of 10 or greater is defined as depression. In the Chinese validation study, the PHQ-9 was reported to have acceptable internal consistency ($\alpha = 0.90$) (68). In the current study, the Cronbach's alpha of the EPDS was 0.899, suggesting the internal consistency of the PHQ-9 was adequate. Research has shown that the PHQ-9 is strongly correlated with other measures of mental health and well-being (69). Its use in caregiving contexts has revealed significant associations between caregiver mental health and the burden they experience.

2.3.4 Fatigue Severity Scale (FSS)

The Fatigue Severity Scale (FSS) is a self-administered questionnaire measuring fatigue (70). It includes nine statements rated on a 7-point scale. The total score is calculated as the mean of the responses to the nine statements (71). A score of 4 or higher indicates a moderate to high level of fatigue (72). In the Chinese validation study, the FSS was found to have a very good internal reliability ($\alpha = 0.93$) (73). In this study, the Cronbach's alpha of the FSS was 0.929. Previous studies have demonstrated a strong correlation between the FSS and other measures of health-related quality of life and psychological well-being (74, 75). It has proven particularly valuable in evaluating fatigue among caregivers, providing insights into how fatigue influences their overall burden and caregiving experiences.

2.4 Statistical analysis

Data were analyzed by SPSS 21.0 software using descriptive and inferential statistics. The sample size of this study was less than 200, so the normality test was judged using Kurtosis and Skewness, with Kurtosis ≤ 3 and Skewness ≤ 10 indicating that the data basically conformed to a normal distribution. Mean and standard deviation (SD) were reported for continuous variables that conformed to a normal distribution, and Median and interquartile spacing were reported for continuous variables that conformed to a non-normal distribution. Number and percentage were reported for categorical variables.

A stepwise model in multiple linear regression is a systematic method for selecting a subset of predictor variables to include in a regression model. This approach is particularly useful when dealing with numerous potential predictors, allowing researchers to identify the most significant ones while avoiding overfilling. The stepwise regression process can be conducted in several ways, including forward selection, backward elimination, and bidirectional elimination. (i) Start with no predictors in the model. (ii) Add the predictor that has the lowest *p*-value (indicating the strongest relationship with the dependent variable) to the model. (iii) Continue adding predictors one by one, each time selecting the one with the lowest *p*-value until no remaining predictors meet the entry criterion. Criteria for entry: A common criterion is a significance level (alpha) such as 0.05. If the *p*-value for a predictor is less than 0.05, it is added to the model.

In the current study, predictor variables include the relationship to the child, age, education level, occupation, marital status, average caring time and family systems, the score of the PHQ-9 and FSS, the response variable is the score of CBI, and the stepwise regression process was conducted by forward selection. The multiple linear regression was undertaken to evaluate the statistical significance of the effect of demographic factors, depression, and fatigue on caregiver burden by forward selection of the stepwise method. The statistically significant value was *p*-value <0.05. The assignment of predictor variables is in Table 1.

3 Results

A total of 165 parents were included in this study. Regarding the age of participants, 32 (19.4%) were ≦30 years, 113 (68.5%) were 31–40 years, and 20 (12.1%) were ≧41 years. Most of the participants were child's mothers ($n = 129$, 78.2%). Other demographic characteristics of participants are displayed in Table 2.

The result indicated that the mean and SD of the CBI score was 42.18 ± 18.79. Among the five domains, time-dependent burden scored the highest (14.64 ± 4.91), followed by developmental burden (12.38 ± 5.78), while emotional burden scored the lowest (2.79 ± 3.36).

The scores of total CBI and domains were positively correlated with PHQ-9 and FSS (*r* ranging from 0.420 to 0.630 and 0.250 to 0.447, respectively, all $p < 0.001$). The mean and standard deviations of the variables under the study and their Spearman correlations are given in Table 3. The total scores of the PHQ-9, which measures levels of depression, exhibited a positive correlation with the scores from the Caregiver Burden Inventory. This indicates that higher levels of depressive symptoms in individuals, as assessed by the PHQ-9, are associated with higher caregiver burden. In other words, caregivers who reported more significant depressive symptoms also tended to experience greater feelings of burden related to their caregiving responsibilities.

The total scores on the FSS, which assesses the severity of fatigue and its impact on daily functioning, exhibited a positive correlation with the scores from the CBI. This finding indicates that higher levels of fatigue reported by caregivers are associated with increased feelings

TABLE 1 The assignment of predictor variables.

Predictors variables	The assignment of variables
Age (years)	"≦30" = 1; "31–40" = 2; "≧41" = 3
Relationship to child	Father = 1; Mother = 2
Educational level	Primary school and below (≤6 years) = 1
	Junior high school (7–9 years) = 2
	Senior high school (10–12 years) = 3
	University (13–16 years) = 4
	Master and above (≥17 years) = 5
Occupation	Employed = 1; Unemployed = 2
Marital status	Married = 1; Divorced = 2
Average caring time (hours/day)	"<4" = 1; "4–8" = 2; ">8" = 3
Family systems	Nuclear family = 1; Stem family = 2; Joint family = 3; Single parent family = 4
Age of child (years)	"<3" = 1; "3–6" = 2; ">6" = 3
Sex of child	Boy = 1; Girl = 2
Types of Cerebral Palsy	Spastic hemiplegia = 1; Spastic diplegia = 2; Spastic tetraplegia = 3; Ataxia = 4; Dyskinetic = 5; Mixed types = 6
GMFCS	I level = 1; II level = 2; III level = 3; IV level = 4; V level = 5
Combined visual impairment	Yes = 1; No = 2
Concomitant epilepsy	Yes = 1; No = 2

GMFCS, Gross Motor Function Classification System.

of burden related to their caregiving roles. The positive correlation likely extends to specific domains of both the FSS and the CBI. This means that not only the overall fatigue levels but also particular aspects of fatigue (such as physical fatigue, mental fatigue, or the impact of fatigue on daily activities) may correlate with specific dimensions of caregiver burden (such as emotional, physical, or social burden). For instance, caregivers reporting significant physical fatigue may also experience heightened emotional or role-related burdens in their caregiving tasks.

The multiple linear regressions analysis revealed that having a low level of education, long caring time, having a child with combined visual impairment, and higher scores of PHQ-9 and FSS pertained to higher scores of CBI (adjusted $R^2 = 0.464$, $F = 29.357$, $p < 0.001$). The results of the multiple linear regression analysis for the factors related to the caregiver burden of parents of children with cerebral palsy are presented in Table 4. Caregivers with a lower level of educational attainment were found to report higher levels of caregiver burden. This suggests that educational background may influence coping strategies and resources available to caregivers. The duration of caregiving is significantly linked to increased caregiver burden. Those who have been providing care for a longer period tend to experience greater levels of stress and exhaustion, likely due to the cumulative effects of long-term caregiving responsibilities. Caregivers of children who have combined visual impairment reported higher burden levels. This may reflect the unique challenges and demands associated with caring for a child with such impairments, which may require more intensive support and resources. Higher scores on PHQ-9 indicate greater depressive symptoms among caregivers, which are associated with increased feelings of burden. This suggests that as caregivers experience more depressive symptoms, their perceived burden also rises. Higher scores on the FSS were also significantly related to higher CBI scores, indicating that greater fatigue levels experienced by caregivers correlate with an increased sense of burden. This may be due to the physical and emotional exhaustion that caregiving can entail.

4 Discussion

To the best of our knowledge, the current study presented the first quantitative research on caregiver burden related to parents of children with cerebral palsy in southwest China. This cross-sectional study was conducted to determine the level of care burden and its related factors in the parents of children with cerebral palsy. The finding of the study detected that the score of care burden in half of parents was of moderate level. In line with the studies conducted in South Africa (76), Spain (50), Turkey (36, 52), Nigeria (49), Poland (48), and Brazil (77), most of the parents of children with cerebral palsy endured moderate to severe care burden and various challenges on account of restricted access to alternative caring resources and the fear for children's development. This high caregiver burden among parents of children with cerebral palsy calls for attention and action by healthcare institutions and disabled persons' federations in China.

According to the result of this study, among of five domains of CBI, time-dependent burden received the highest score with a mean ± SD of 14.64 ± 4.9, followed by developmental burden scoring 12.38 ± 5.78 in comparison to other aspects. The heavy time-dependent burden can deplete the caregiver's energy and increase negative emotions (78). The present study illustrated that parents of children with cerebral palsy tend to feel time-related pressure and restriction of their social and cultural activities due to multiple additional care tasks, which is consistent with previous studies (79, 80). The result of multiple linear regression analysis found that the level of education, caring time, visual impairment, the degree of depression, and fatigue remained statistically significant correlates of the caregiving burden.

According to a study conducted in Nigeria (77), parents of children with cerebral palsy who have higher education suffer less caring burden than those with low or no education. Barros et al. (77) pointed out that caregivers with lower educational levels are more likely to fall into poverty and undergo stress. Parents may face societal stigma regarding their child's disability, which can be more pronounced for those who are less educated (66). This stigma can manifest as judgment or lack of understanding from others, leading to feelings of shame and isolation (37). Moreover, parents may internalize societal stigma, leading to feelings of worthlessness or failure in their parenting role (81). This can contribute to a negative self-image and increased caregiver burden (42). Addressing these issues requires targeted support, education, and community awareness to help alleviate the burdens faced by these families, fostering a more inclusive and understanding environment for parents and children with disabilities (82). By providing education and resources, communities can help reduce stigma and empower

TABLE 2 Demographic characteristics of participants and the score of CBI (n = 165).

Variables	Number (n)	Percentage (%)	The score of CBI (Mean ± SD)	Kurtosis	Skewness
Age (years)					
≦30	32	19.4	42.78 ± 15.61	−0.874	0.057
31–40	113	68.5	41.20 ± 20.03	−0.914	−0.177
≧41	20	12.1	46.80 ± 15.99	−0.492	−0.053
Relationship to child					
Father	36	21.8	37.67 ± 19.60	−0.814	0.168
Mother	129	78.2	43.44 ± 18.43	−0.654	−0.286
Educational level					
Primary school and below (≤6 years)	10	6.1	58.50 ± 8.95	−2.200	−0.099
Junior high school (7–9 years)	22	13.3	42.55 ± 19.28	−0.831	0.357
Senior high school (10–12 years)	31	18.8	39.68 ± 20.13	−0.949	−0.380
University (13–16 years)	89	53.9	42.30 ± 17.93	−0.625	−0.161
Master and above (≥17 years)	13	7.9	34.15 ± 20.81	−0.634	0.439
Occupation					
Employed	75	45.5	46.04 ± 16.83	−0.771	−0.183
Unemployed	90	54.5	38.97 ± 19.80	−0.885	−0.073
Marital status					
Married	162	98.2	41.83 ± 18.70	−0.801	−0.189
Divorced	3	1.8	61.33 ± 15.37	0.000	1.658
Average caring time (hours/day)					
<4	35	21.2	36.25 ± 18.38	−0.638	0.021
4–8	34	20.6	31.94 ± 20.34	−0.803	0.336
>8	96	58.2	47.97 ± 16.13	−0.849	−0.197
Family systems					
Nuclear family	58	35.1	40.19 ± 18.27	−0.67	−0.236
Stem family	61	37	43.97 ± 18.80	−0.589	−0.289
Joint family	33	20	39.45 ± 19.46	−1.157	0.061
Single parent family	13	7.9	49.62 ± 18.58	−0.025	−0.39
Age of child (years)					
<3	82	49.7	42.45 ± 15.74	−0.118	0.078
3–6	54	32.7	42.54 ± 20.79	−1.215	−0.365
>6	29	17.6	40.76 ± 23.01	−1.211	−0.122
Sex of child					
Boy	99	60	43.90 ± 19.33	−0.816	−0.266
Girl	66	40	39.59 ± 17.75	−0.621	−0.140
Types of Cerebral Palsy					
Spastic hemiplegia	83	50.3	36.63 ± 19.00	−0.754	−0.186
Spastic diplegia	25	15.2	43.92 ± 16.80	−0.378	−0.500
Spastic tetraplegia	31	18.8	49.48 ± 15.53	−0.699	0.097
Ataxia	8	4.8	53.00 ± 19.00	0.000	0.000

(Continued)

TABLE 2 (Continued)

Variables	Number (*n*)	Percentage (%)	The score of CBI (Mean ± SD)	Kurtosis	Skewness
Dyskinetic	2	1.2	49.00 ± 26.87	−1.536	−0.054
Mixed types	16	9.7	47.88 ± 18.61	−0.282	−0.968
GMFCS					
I level	70	42.4	36.44 ± 20.27	−0.940	0.155
II level	39	23.6	43.00 ± 17.10	−0.299	−0.154
III level	28	17	48.64 ± 15.78	−1.192	−0.088
IV level	14	8.5	43.64 ± 17.62	−0.419	−0.496
V level	14	8.5	54.21 ± 12.70	0.154	−0.262
Combined visual impairment					
Yes	69	41.8	47.23 ± 18.65	−0.357	−0.607
No	96	58.2	38.55 ± 18.13	−0.645	0.059
Concomitant epilepsy					
Yes	8	21.8	41.00 ± 15.50	0.423	−0.953
No	157	78.2	42.24 ± 18.98	−0.8	−0.181

CBI, Caregiver Burden Inventory; SD, Standard Deviation; GMFCS, Gross Motor Function Classification System.

TABLE 3 The mean and standard deviations of the variables under the study and their correlations (n = 165).

Variables	Mean	SD	1	2	3	4	5	6	7	8
1. PHQ-9	7.98	5.56	1							
2. FSS	4.21	1.25	0.513	1						
3. Time-dependent burden	14.64	4.91	0.420	0.250	1					
4. Developmental burden	12.38	5.78	0.533	0.428	0.705	1				
5. Physical burden	7.83	5.00	0.617	0.447	0.608	0.843	1			
6. Social burden	4.54	3.82	0.576	0.406	0.404	0.619	0.676	1		
7. Emotional burden	2.79	3.36	0.474	0.309	0.334	0.519	0.519	0.667	1	
8. Total burden of CBI	42.18	18.79	0.630	0.461	0.744	0.923	0.906	0.783	0.689	1

$p < 0.001$ for all correlations; SD, Standard Deviation; PHQ-9, Patient Health Questionaire-9; FSS, Fatigue Severity Scale; CBI, Caregiver Burden Inventory.

TABLE 4 The results of the multiple linear regressions analysis for the factors related to caregiver burden of parents of children with CP (n = 165).

Variables	B	SE	β	*t*-value	*p*-value
Constant	26.879	7.727	–	3.479	0.001
Education level	−2.231	1.121	−0.121	−1.991	0.048*
Average caring time (hours/day)	3.583	1.392	0.155	2.574	0.011*
Combined visual impairment	−6.133	2.214	−0.162	−2.77	0.006*
PHQ-9	1.618	0.23	0.479	7.029	<0.001**
FSS	2.68	1.016	0.179	2.638	0.009*

B, Unstandardized Coefficient; SE, Standard Error; β, Standardized Coefficient; $R^2 = 0.480$, adjusted $R^2 = 0.464$, $F = 29.357$, *, $p \leq 0.05$, **, $p \leq 0.001$.

parents, ultimately improving their quality of life and the care they provide (83). Higher parental schooling, on the one hand, may enable them to search and acquire information and support resources more efficiently and on the other hand, foster self-efficacy in managing their children's health issues and formulating effective plans to cope with their stressors (84). There is compelling evidence that education is one of the most important sources for caregivers to acquire competency, affecting the perception of stress, resilience, and

psychological flexibility (85). For this reason, caregiver skill training was developed by the World Health Organization (WHO) aiming to facilitate access to parenting skills and strategies for caregivers of children with developmental delays or disabilities (86–89).

We found that the parents with longer caring time perceived a higher level of burden. This finding supports previous research that the length of caregiving is associated with caregiver burden. In terms of time cost, Park et al. (80) reported that parents of children with cerebral palsy spend about 14–15 h per day to support their children in Korea. A study conducted in Australia revealed a significant positive correlation between psychological problems and caring time (90). In the present study, the caring time on an average day was also fairly long, with 58.2% of parents spending more than 8 h per day devoted to caring for their children. On one hand, increased time allocated by the parents for their children might require one of the parents to quit work to care for the child, which adds a financial burden to the families. On the other hand, parents with longer caring time might feel more burnout and pain, which, in turn, enhances the care burden experienced as well (36).

The results of the current study demonstrated that the caregiver burden was higher in parents of children with visual impairment, which is consistent with previous research (44, 91). This might be attributed to the fact that visually impaired children need additional care for daily tasks due to their diminished independent living skills and increased likelihood of injury (44). Most of the qualitative studies conducted with caregivers of children with cerebral palsy documented that most caregivers worry about their children's future (42). Thus, parents of visually impaired children with cerebral palsy are more likely to feel tiredness and pressure.

In keeping with the views in the literature (48, 92), it was observed that caregiving burden was significantly affected by depression and fatigue. Caregivers experiencing higher levels of depression may feel more overwhelmed by their responsibilities, which can create a cycle of increased stress and emotional strain (93). This highlights the importance of addressing the mental health needs of caregivers, as their psychological well-being is closely linked to their experience of burden (94). This has major clinical implications for medical practitioners and nurses in being able to identify parents at risk for burden. The current study suggests a significant relationship between the experience of fatigue and the perception of caregiver burden. Caregivers who report higher levels of fatigue may feel less capable or more drained in their caregiving roles, contributing to a greater sense of burden (95). This can create a vicious cycle where fatigue exacerbates feelings of burden, potentially leading to further fatigue and emotional distress (96). This highlights the necessity for interventions and support systems aimed at addressing fatigue among caregivers, which could, in turn, alleviate their burden and improve their overall quality of life (97). Parents with a greater number of depressive and fatigue symptoms may require more individualized support and interventions, such as problem-solving therapy and group educational intervention (98, 99).

The study contributes to providing existing evidence on caregiver burden in a Chinese context. However, there are a few limitations of this study. Firstly, the nature of cross-sectional study prevents the inference of causal relationships. Secondly, the overall multiple regression models explained only up to 46.4% of the variance in caregiver burden, which was relatively low, indicating the need for more extensive research on the correlates of caregiver burden. Third, limitation of quantitative approaches, particularly regarding the inability to capture the nuanced experiences and specific emotions that parents may face. While our quantitative design allowed for the measurement of broader trends and relationships, it did not provide the depth of understanding that qualitative methods offer.

5 Conclusion

In conclusion, this study showed that most parents of children with cerebral palsy experience moderate to severe caregiver burden, the time-dependent burden being the heaviest compared with other types of burden. A moderate burden may refer to caregivers who feel overwhelmed but can still manage their responsibilities, while a severe burden indicates that they are experiencing significant distress and may struggle to cope with their caregiving duties. The key predictors of caregiver burden include the level of education, caring time, children with visual impairment, and the degree of depression and fatigue. These findings underscore the multifaceted nature of caregiver stress and the importance of addressing these factors to support caregivers effectively. The current findings may prove useful for providing more effective family-oriented support programs for parents of children with cerebral palsy. Taking into account the factors linked to caregiver burden, further studies should examine the burden of care in different societies with diverse cultures.

Data availability statement

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

Ethics statement

The studies involving humans were approved by The Ethics Committee approval of the West China Second University Hospital, Sichuan University (Approval ID: 2021/218). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

XZ: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. CW: Writing – review & editing.

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Conflict of interest

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

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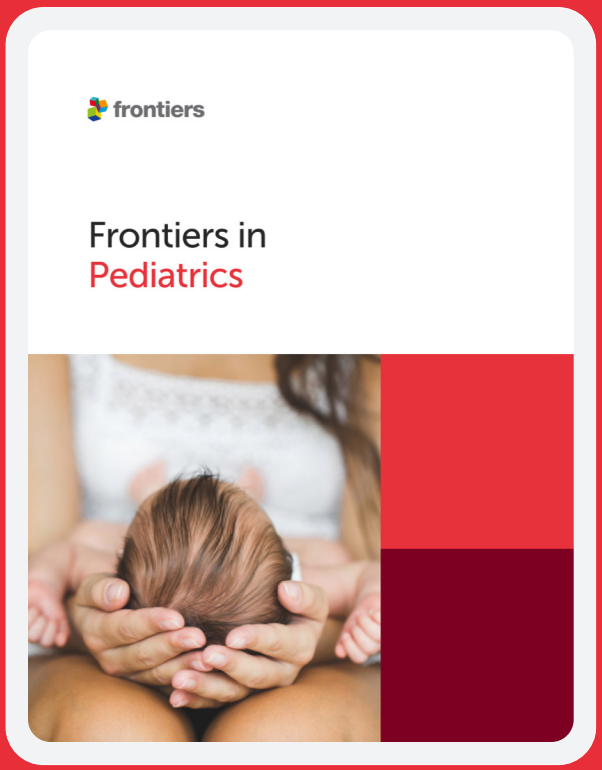

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