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Families and functioning in childhood and adolescence

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Families and functioning in childhood and adolescence

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EDITORIAL
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Editorial: Families and Functioning in Childhood and Adolescence

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Editorial on Research Topic

Families and Functioning in Childhood and Adolescence

In WHO's International Classification of Functioning, Disability, and Health (ICF), "functioning" refers to the sum of functions and structures of the body and mind, the actions people perform, and the execution of activities when participating in diverse life situations. These interact with personal and environmental factors. Functioning in childhood and youth is highly dependent on the family. Every family is different, and the concept of family has changed over time and is diverse in different geographic regions of the world. According to the American Academy of Family Physicians: "A family is a group of individuals with a continuing legal, genetic, and/or emotional relationship. Society relies on the family group to provide for the economic and protective needs of individuals, especially those who are vulnerable." (1).

In the field of child development disciplines (e.g., early intervention, developmental paediatrics, social paediatrics, paediatric rehabilitation, child and youth mental health, etc.) the so-called "main complaints" are brought to the attention of professionals by family members or caregivers. We see and support families who live and struggle with their children and youth. Supporting and encouraging the people with whom a child lives and interacts and improving other environmental factors promotes a child's activity and participation opportunities and improves the child's quality of life. It is a person-centred approach taking into consideration a child's life situation. On the other hand, we know that goals defined by parents would not necessarily match with those a child would describe as meaningful for them (2).

Authors from around the world followed our invitation to publish their ideas and research findings about how to engage and consider family aspects in working with children and adolescents. Those ideas share a common approach in seeing a person within their environment and life situation (3). We are excited to share an extremely diverse, intriguing, and thought-provoking collection of contributions. Embracing a family-centred attitude, some of the articles were co-written with families (parents and youth). This intimate collaboration reflects a paradigm shift described by Nowotny et al. (4) in their book "Re-thinking Science": instead of science speaking to society, we are living in a time where society speaks to science. New knowledge and insights are co-created and developed within meaningful contexts. The science ivory tower is open to the agora of the people for an inclusive conversation.

"Scientists listening to families" is the topic of two articles that describe the perspectives of siblings and fathers, two groups often easily overlooked when caring for children with complex or chronic health conditions (Nguyen et al.; Ogourtsova et al.). Another easily overlooked aspect is that of non-traditional family constellations such as LGBTQ + parents and polygamous and polyamorous families. Phoenix et al. describe how clinicians and service providers can easily create barriers for them to access services by perpetrating systemic patterns of racism, sexism, and ableism. In this vein also belongs the reflection of Reitzel et al. on how an intersectional lens between personal and

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environmental factors can help us identify the risk for discrimination by expanding the use of the ICF.

With relation to service structures, the importance of family-centred care and how it influences functioning of children and adolescents is described by Rosenbaum who also contributes extensively in disseminating those ideas around the world, as the example from Brazil in this collection demonstrates (Airoldi et al.).

One of the important contributors to child and adolescent functioning is the perceived parental social support as described by Weiss et al. following families with autistic children longitudinally. Such support is further compromised the current COVID-19 pandemic for families with children with disabilities impacting their quality of life as shown by Ali et al. with data from Pakistan.

When families are not available, and children or youth are being cared for by child protection services, different aspects and factors need to be considered. Kim et al. demonstrate in their review of longitudinal studies through the "ICF lens" how having a distal and proximal assessment of functioning can help in understanding the different risks and trajectories observed in children in care. This insight is innovative and might help in conceptualizing future longitudinal studies within the ICF framework.

We hope that these ideas stimulate our readers to reflect on their own practice and help them to develop inclusive approaches to engage children and families in clinical practice and research.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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ORIGINAL RESEARCH
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Understanding Stability and Change in Perceived Social Support in Parents of Autistic Children and Adolescents

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Parents of children with autism often have their own support needs. Informal social support can be an important component of managing parenting-related stressors. We know very little about the factors that lead to higher levels of perceived social support or the potential reciprocal relationship social support has with other factors in parents of children with autism. The current longitudinal study examined the reciprocal relations of perceived social support and parent stress and child behavior problems across a 1-year period, using three time points. There was remarkable stability in variables over time. Baseline perceived social support significantly predicted changes in child behavior and parent stress at the 6-month time point, but neither of those variables significantly predicted social support. This study adds to our understanding of social support and clarifies how perceived social support relates to other factors longitudinally.

Keywords: social support, autism, parent stress, behavior problems, longitudinal design

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INTRODUCTION

The benefits of social support are well-documented for parents of individuals with autism [e.g., (1–7)]. Cohen et al. (8) state that social support is "the social resources that persons perceive to be available or that are actually provided to them by non-professionals in the context of both formal support groups and informal helping relationships" (p. 4). *Perceived social support* is the belief that support is adequate or available if needed, reflecting how supported a person feels rather than specific or concrete supports experienced (9). It appears that perceived support can help with a person's well-being regardless of the stressors experienced (10) and may be particularly important within the context of chronic and acute negative life events (11).

Perceived support is consistently linked to well-being in parents, including lower levels of stress (12), depressive symptoms (13), distress (14), and increased self-confidence (15). This pattern is also found in parents of people with autism as well (2), with perceived support being associated with lower mental health problems and greater life satisfaction and general well-being, in both mothers and fathers (4, 16–19). One of the most commonly studied correlates of perceived support is parents' stress (20), broadly defined as the distress, discomfort, or arousal experienced in response to perceived demands.

Child behavior problems is a particularly relevant variable to consider in reference to perceived support for parents of people with autism. Individuals with autism may struggle with high levels of irritability, emotion regulation problems, aggression, or self-injurious behaviors, which regularly require parent support (21). For example, in a study of 1,380 parents of children and adolescents with autism, nearly 70% reported that their child had demonstrated aggression toward caregiver, and half toward non-caregivers (22). There is some research to suggest that caregivers may struggle to mobilize support or be more reluctant to seek support when children with autism show more difficult behaviors. From interviews with 46 parents of children with autism, Ryan (23) described parent reluctance to enter public places and struggles to find social acceptance. Without obvious outward signs of their child's disability, parents often perceived judgement from the community when their child acted out or pushed societal norms. Similarly, Gray's (24) qualitative study involving 33 Australian parents of school-aged children (4–19 years) with autism found that parents withdrew from their social networks in response to perceived stigma and the stressful nature of public encounters. Many parents report heightened feelings of isolation when their child had aggressive or disruptive behavior, suggesting a potential link between child behavior and perceived availability of support. A decade later, Gray (25) interviewed 28 of these families again to examine how coping changes over time. Parents reportedly felt more comfortable engaging in social activities in the community because they perceived their child's behaviors to have improved, but parents also had grown accustomed to the longstanding social restrictions that existed for their families. Cross-section surveys confirm this negative correlation between child behavior problems and social support, found in studies of very young children to late adolescence (3, 18, 26).

While there is substantial cross-sectional information to support the idea of associations, we know little about the directionality of perceived social support. To date, no study has examined stress as a determinant of perceived social support longitudinally for parents of individuals with autism, or considered a bidirectional relationship between stress or child behavior problems and perceived social support. While it is possible that changes in stressors or child functioning leads to changes in perceived support, it is also possible that greater support leads to improved perceptions of stressors and stress-responses. More broadly, there is evidence that these variables show a degree of stability over short periods of time (27–30) and it is an empirical question as to the degree of change and stability that is witnessed in community samples of stress, perceived support, and child behavior problems. Using online survey data collected from 249 parents of school-aged children with autism, the current study assessed the relationships between perceived social support and parent perceived stress and child behavior problems across three time points, within a 1-year period. While we expected stability in terms of child behavior problems, we also considered that their presence would lead to changes in perceived social support over time, and that changes in support would lead to changes in behavior problems. Similarly, though we would see stability in parent stress levels, above this we expected strong relationships with support over time.

METHODS

Participants

Baseline data were available for 249 parents who sufficiently completed an online survey (i.e., at least 75% of survey items) and met all eligibility criteria (described below). At time 2 (6 months after baseline), 194 participants responded. At time 3 (12 months after baseline), there were 180 participants (17 of these participants did not respond at time 2). The study had 163 participants complete all three surveys. The 163 participants who sufficiently completed all three time points were compared to the 86 parents who did not. The two groups did not significantly differ on the main study variables or on family and child characteristics including parent education, household income, child age, child autism symptoms, and child adaptive skills (all $p > 0.05$).

As shown in **Table 1**, parent age ranged from 27 to 62 years ($M = 43.98$, $SD = 6.2$, Median = 44). Participants were primarily mothers (95.6%) and currently married/common law (83.1%). Most parents (81.9%) had graduated college or university. Parents were from suburban (39.9%), urban (39.1%), rural (16.5%), and remote (4.4%) settings across Canada. The children with autism ranged in age from 4 to 18 years ($M = 11.47$, $SD = 3.95$, Median = 11) and most were male (83.1%). Additional child diagnoses from a physician, as reported by parents, included intellectual disability (42.4%), learning disability (37.8%), attention deficit disorder or attention deficit hyperactivity disorder (38.4%), anxiety or depression (37.1%), and behavior or conduct problems (29.0%). Nearly half (45.7%) had at least one chronic health condition, including epilepsy, cerebral palsy, or asthma.

Measures

Demographics

Parents reported their own age, gender, marital status, and income as well as their child's age, gender, and diagnoses.

Autism Symptoms

The Social Communication Questionnaire—Lifetime (SCQ) (31) was used to assess autism symptom severity. The SCQ is an autism symptom screener assessing social and communication behaviors and consists of 40 yes-or-no items. Higher total scores indicate greater autism symptom severity. The SCQ has shown strong internal consistency, as well as good discriminant validity for distinguishing between children with autism and those without (32). In the current study, baseline scores had adequate internal consistency (coefficient $\alpha = 0.82$).

Child Adaptive Behavior

Adaptive behavior was measured as a control variable, using the Waisman Activities of Daily Living Scale (W-ADL) (33). This is a 17-item measure of an individual's independence in performing daily activities (e.g., dressing and undressing or drinking from

TABLE 1 | Parent, household, and child characteristics.

	N (%) or M (SD)
Parent/household variables	
Age ($n = 233$)	43.98 (6.21) Range: 27–64
Gender	
Female	238 (95.6)
Male	10 (4.0)
Transgender	1 (0.4)
Relationship status ($n = 248$)	
Married/common law	210 (83.1)
Single (never married)	10 (4.0)
Separated/divorced	31 (12.5)
Widowed	1 (0.4)
Education level ($n = 248$)	
High school or less	23 (9.2)
Partial college (at least 1 year)	22 (8.9)
College diploma/university undergraduate degree	150 (60.5)
Graduate degree	53 (21.4)
Annual household income after taxes ($n = 244$)	
$45,000 or less	57 (23.4)
$45,000–95,000	105 (43.0)
$95,000 or more	82 (33.6)
Geographical Location ($n = 248$)	
Suburban area	99 (39.9)
Urban area	97 (39.1)
Rural	41 (16.5)
Remote	11 (4.4)
Child variables	
Age	11.47 (3.95) Range: 4–18
Gender	
Female	41 (16.5)
Male	207 (83.1)
Transgender	1 (0.4)
Born outside of Canada	12 (4.8)
Activities of daily living skills (W-ADL)	16.69 (7.11) Range: 0–33
Autism Symptoms (SCQ)	22.17 (6.34) Range: 11–38

N = 249.

a cup). Item responses are given using a three-point Likert-type scale, with 0 = *Does not do at all* and 2 = *Independent or does on own*. Total scores range from 0 to 34. The WADL has been used with parents of children with intellectual disabilities [e.g., (34)] and with adolescents and adults with autism and no intellectual disability (35). Maenner et al. (33) report good internal consistency and strong validity, as the scale is highly correlated with other measures of adaptive functioning. In the current study, baseline scores had good internal consistency (coefficient $\alpha = 0.92$).

Child Behavior Problems

Child behavior problems were assessed using the Strengths and Difficulties Questionnaire [SDQ; (36)]. The 25 items assess prosocial behavior, peer relationship problems, conduct problems, hyperactivity, and emotional symptoms. Each item is scored using a 3-point scale (*not true*, *somewhat*, and *certainly true*) and a total difficulties score is calculated by summing the four problem behavior subscales. Example items include "generally liked by other children," "easily distracted, concentration wanders," and "often loses temper." The scale is meant to serve as a brief behavioral screener and is often used in research involving parents of children with developmental disabilities or autism [e.g., (37, 38)]. In the present study, prosocial behavior and peer subscales were not used because they represent areas of functioning represented in the diagnostic criteria for autism, consistent with other studies [e.g., (21)]. The SDQ has shown good internal consistency, test-retest reliability, and validity for parents of typically developing children (39) and internal consistency has been high in a sample of parents of children with autism (0.97) (38). For the current study, coefficient $\alpha = 0.78$ for baseline total difficulties (sum of conduct problems, hyperactivity and emotional symptoms).

Parent Stress

The Stress subscale from the Depression Anxiety and Stress Scale (DASS-42) (40) is a 14-item scale assessing global perceptions of stress. The stress subscale measures the extent to which individuals had difficulty relaxing, feelings of nervousness, agitation, intolerance, impatience, or irritability in the last week. Item responses are given on a four-point Likert-type scale from 0 (*did not apply to me at all*) to 3 (*applied to me very much or most of the time*), where higher scores suggest more perceived stress. Example items include "I found it difficult to tolerate interruptions to what I was doing" and "I was in a state of nervous tension." The scale has shown acceptable reliability for parents of children with developmental disabilities or autism [e.g., (41)], with coefficient α of 0.85 in a similar study sample (42). Good validity has been demonstrated with a sample of adult psychiatric patients (43) and a non-clinical sample (44). In the current study, baseline scores had good internal consistency (coefficient $\alpha = 0.94$).

Perceived Social Support

Perceived social support was measured with the Social Provisions Scale (45). The scale provides a summary score of global perceived availability of social support. The 24 items are scored using a four-point Likert-type scale ranging from *strongly disagree* to *strongly agree*, with higher scores suggesting greater perceptions of support. Example items include "I feel part of a group of people who share my attitudes and beliefs," "there are people I can count on in an emergency," and "there is someone I could talk to about important decisions in my life." The scale had excellent internal consistency in a large-scale study of its psychometric properties (coefficient $\alpha = 0.92$) and good convergent and divergent validity (45). The scale has also shown good reliability in studies involving parents of children with behavior difficulties (46) and autism (47). In the current study, baseline scores had good internal consistency, $\alpha = 0.94$.

Procedure

Following approval from University's Research Ethics Board, parents of individuals with autism were recruited through postings on the Canadian autism websites, community organizations, and through an ongoing research database available through the primary researcher's lab. A link to the online consent form and survey was provided and parents were invited to contact the researcher by email or phone to request a paper survey. After parents completed the initial survey, they were invited to complete follow-up surveys 6 and 12 months later.

To be eligible for this study, participants were required to have a school-aged child (between 4 and 18 years of age) with a confirmed diagnosis of an Autism Spectrum Disorder and be able to complete the survey in English. Autism diagnosis was confirmed in two ways. First, the parent confirmed that a professional with the capacity to diagnose provided the child with an autism-related diagnosis (selecting one of the of the following: psychologist, psychiatrist, developmental pediatrician, general pediatrician, family doctor, nurse practitioner, multidisciplinary or developmental team, genetic testing, neurologist) and provided the date of diagnosis. Second, the parent-reported score on the Social Communication Questionnaire—Lifetime (SCQ) (31) was above a pre-specified cut-off score of 11, indicating a possible autism diagnosis (48).

Data Analysis Plan

Two separate autoregressive cross-lagged path models were calculated, allowing for individual examination of stress and child behavior problems with perceived social support across the three time points. This type of statistical model is used to examine transactional relationships between variables and has recently been used in the field of autism research [e.g., (35, 38, 49, 50)]. The model allows for examination of the directionality of effects between two variables measured over time while also considering auto-regression, which is variable stability across time points. Model fit was assessed using a series of common fit statistics such as comparative fit index (CFI), root mean square error of approximation (RMSEA), and Tucker-Lewis index (TLI). The individual parameter estimates pertaining to the cross-lagged effects were subsequently interpreted. Robust maximum likelihood estimation (MLR) (51) was used to account for the possibility of multivariate non-normality and for its effectiveness in dealing with missing data. Demographic variables that showed a significant association with model variables at the bivariate level were included as control variables, to account for many additional stressors that could inadvertently influence the presence of stress, child behavior problems and support, ultimately representing more conservative findings.

RESULTS

As shown in **Table 2**, the means and within-variable correlations indicated considerable stability. Further, at Time 1, perceived support was correlated with child adaptive behavior level ($r = 0.22, p < 0.001$), child autism symptom severity ($r = -0.18, p = 0.003$), the presence of at least one chronic health condition [$t_{(244)} = 2.0, p = 0.04$], parent education level ($r = 0.27, p < 0.001$), and household income level ($r = 0.22, p = 0.001$). Child behavior problems was associated with child autism symptom severity ($r = 0.15, p = 0.02$), and parent education level ($r = -0.18, p = 0.005$). Stress was associated with child adaptive behavior ($r = -0.19, p = 0.003$), child autism symptom severity ($r = 0.15, p = 0.02$), and parent education level ($r = -0.16, p = 0.01$). Higher perceived social support was significantly related to lower levels of parent stress ($r = -0.44$ $p < 0.001$) and child behavior problems ($r = -0.17, p = 0.01$). Given this pattern, household income, parent education, presence of child chronic health conditions, child adaptive behavior level, and child autism symptom severity were entered as control variables in both path models.

Is There a Reciprocal Relation Between Perceived Social Support and Child Behavior Problems, While Controlling for Continuity Over Time for Both Variables?

Initial model fit for this model was poor (CFI = 0.86; TLI = 0.56; RMSEA = 0.17; SRM $r = 0.09$). Residual correlations showed strong autoregressive relationships between variables at Time 1 and Time 3, and modification indices suggested that adding direct paths between T1 and T3 would substantially improve the model fit. The adjusted model fit the data well (CFI = 1.0; TLI = 1.0; RMSEA = 0.00; SRM $r = 0.01$). See **Table 3** for unstandardized estimates and **Figure 1** for the corresponding path diagram with standardized parameter estimates.

TABLE 2 | Descriptive and within-variable correlations of main study variables across time points.

	Baseline M (SD)	6 months M (SD)	12 months M (SD)	T1-T2 *r*	T2-T3 *r*	T1-T3 *r*
Perceived social support (SPS)	75.06 (11.85)	76.33 (11.46)	74.46 (12.29)	0.77*	0.78*	0.80*
Stress (DASS)	15.46 (8.91)	15.75 (9.43)	14.55 (8.90)	0.60*	0.61*	0.65*
Child behavior (SDQ)	12.86 (5.09)	13.55 (4.72)	13.17 (4.90)	0.65*	0.78*	0.62*

*$p < 0.05$.

TABLE 3 | Unstandardized estimates of the relationships between perceived social support and child behavior problems.

	Estimate (SE)	*p*
SS 6 months		
SS baseline	0.72 (0.05)	<0.001
Behavior baseline	0.08 (0.10)	0.44
Education	0.40 (0.68)	0.55
Household income	−0.07 (0.19)	0.70
Child health condition	−0.82 (1.04)	0.43
Autism symptoms	−0.18 (0.08)	0.02
Adaptive skills	0.10 (0.08)	0.21
Behavior *6 months*		
Behavior baseline	0.58 (0.06)	<0.001
SS baseline	−0.06 (0.02)	0.02
Education	0.04 (0.30)	0.97
Household income	−0.03 (0.09)	0.71
Child health condition	0.19 (0.53)	0.67
Autism symptoms	−0.01 (0.04)	0.95
Adaptive skills	−0.01 (0.04)	0.64
SS 12 months		
SS 6 months	0.43 (0.07)	<0.001
SS baseline	0.46 (0.07)	<0.001
Behavior 6 months	−0.24 (0.12)	0.05
Behavior *12 months*		
Behavior 6 months	0.70 (0.07)	<0.001
Behavior baseline	0.22 (0.06)	<0.001
SS 6 months	−0.01 (0.02)	0.72

SS = perceived social support.

There were significant autoregressive effects for both perceived social support and child behavior problems, indicating that the prior levels of either variable were strongly related to the same variable's subsequent levels. Specifically, baseline to 6-month social support ($b = 0.72$, $p < 0.001$), 6- to 12-month social support ($b = 0.43$, $p < 0.001$), baseline to 6-month child behavior ($b = 0.58$, $p<0.001$), and 6- to 12-month behavior ($b = 0.70$, $p < 0.001$) were all significant autoregressive effects. Cross-lagged effects showed baseline social support significantly predicted child behavior problems at 6 months ($b = -0.06$, $p = 0.02$), but baseline behavior did not significantly predict 6-month social support. There were no significant cross-lagged paths from 6 to 12 months.

Is There a Reciprocal Relation Between Perceived Social Support and Parent Stress, While Controlling for Continuity Over Time for Both Variables?

The initial planned model with perceived social support and parent stress had an inadequate fit to the data (CFI = 0.84; TLI = 0.70; RMSEA = 0.14; SRM $r = 0.10$). Based on residual correlations and modification indices, direct paths from social support at time 1 to time 3 and from stress at time 1 to time 3 were added to the model. This modification improved model fit such that the adjusted model fit the data well (CFI = 1.0; TLI = 0.99; RMSEA = 0.03; SRM $r = 0.02$). Unstandardized results for this model are reported in **Table 4**. As shown in **Figure 2**, both perceived social support and stress were stable over time. Specifically, autoregressive coefficients from baseline to 6-month social support ($b = 0.71$, $p < 0.001$), 6- to 12-month support ($b = 0.47$, $p < 0.001$), baseline to 6-month stress ($b = 0.57$, $p < 0.001$), and 6- to 12-month stress ($b = 0.33$, $p < 0.001$) were all significant.

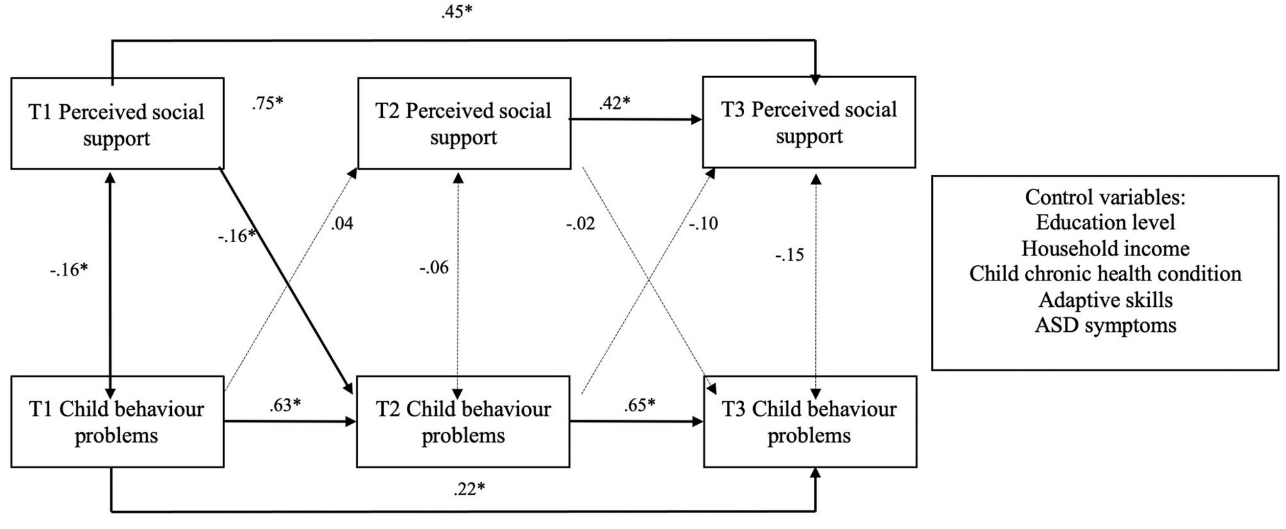

FIGURE 1 | Standardized coefficients of the relationships between perceived social support and child behavior problems across three time points. T1 = baseline; T2 = 6 months; T3 = 12 months; Dotted lines represent non-significant associations; *$p < 0.05$.

TABLE 4 | Unstandardized estimates of the relationships between perceived social support and stress.

	Estimate (SE)	*p*
SS 6 months		
SS baseline	0.71 (0.05)	<0.001
Stress baseline	−0.04 (0.06)	0.43
Education	0.29 (0.65)	0.66
Household income	−0.04 (0.19)	0.81
Child health condition	−0.67 (1.04)	0.52
Autism symptoms	−0.17 (0.08)	0.03
Adaptive skills	0.09 (0.08)	0.29
Stress 6 months		
Stress baseline	0.57 (0.07)	<0.001
SS baseline	−0.15 (0.05)	0.006
Education	1.76 (0.72)	0.02
Household income	−0.44 (0.19)	0.02
Child health condition	−0.01 (1.14)	0.99
Autism symptoms	−0.10 (0.09)	0.31
Adaptive skills	−0.06 (0.08)	0.48
SS 12 months		
SS 6 months	0.46 (0.08)	<0.001
SS baseline	0.44 (0.07)	<0.001
Stress 6 months	−0.05 (0.06)	0.36
Stress 12 months		
Stress 6 months	0.33 (0.09)	<0.001
Stress baseline	0.41 (0.08)	<0.001
SS 6 months	−0.01 (0.04)	0.77

SS = perceived social support.

The cross-lagged path from baseline social support to stress at 6 months was significant ($b = -0.15$, $p = 0.006$), indicating that higher baseline social support is associated with lower levels of stress at 6 months. All other cross-lagged paths were non-significant.

DISCUSSION

Results shed light on how perceived support uniquely relates to parent stress and child problem behavior. As expected, there was remarkable stability in our three variables over a 1-year period. In community samples, it is common to find that without major treatment initiatives, many of the stressors associated with chronic neurodevelopmental conditions are themselves chronic, adding to the level of short term relative stability in the presence of behavior problems, which may take longer to change than a 1-year period (52). Stress itself has been described as a state that is associated with stable personality traits, such as neuroticism (53), it is likely that these parent characteristics also inserted a degree of relative consistency across respondents. Given that we can expect little change in stressors, stress, and support within a year, it is important to consider ensuring that families have access to interventions to address their needs in a timely manner to foster greater positive change.

Coupled with the fact that this stability was confirmed over a 1-year period, and that we controlled for many additional potential confounding variables, including parent education level and family household income, child health conditions, and the level of children's adaptive and autism symptoms, it is perhaps unsurprising that the degree of cross-lagged variance accounted for in our models was small though significant in the first 6-month period. Cross-lagged models are unique in that they

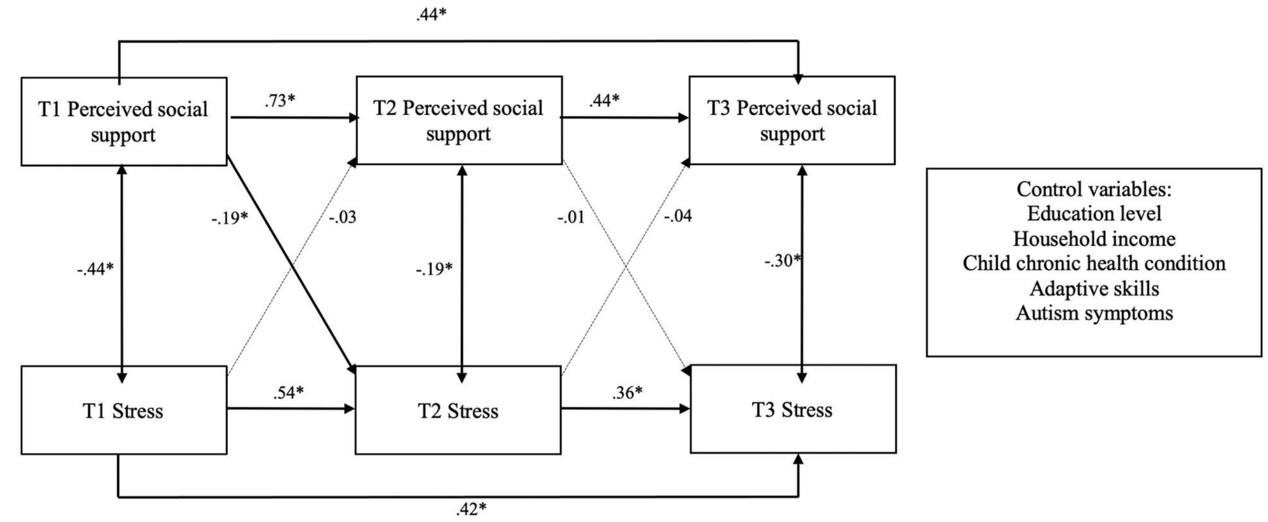

FIGURE 2 | Standardized coefficients of the relationships between perceived social support and stress across three time points. T1 = baseline; T2 = 6 months; T3 = 12 months; Dotted lines represent non-significant associations; *$p < 0.05$.

control for variable stability across time points and are better equipped to assess reciprocal relationships. This methodology has been used in the autism research field to elucidate the reciprocal relationships among expressed emotion and behavior problems in adults (54), child anxiety and over-responsivity (49), adolescent behavioral development and vocational engagement (35), and child behavior and parent well-being (38, 55). In the current analysis, baseline perceived social support predicted 6-month child behavior and 6-month stress, but neither of the latter variables predicted subsequent social support. Greater within-subject variability over time may have resulted in more observations of cross-lagged effects.

The existing research framing child behavior problems as a determinant of perceived social support posits that caregivers may struggle to mobilize supports or are more reluctant to seek support when their children have more difficult behaviors [e.g., (3)]. This pattern was evident in our bivariate correlation analyses, as perceived social support was negatively associated with increased child behavior problems. However, results did not confirm this pattern longitudinally. Specifically, baseline perceived support significantly predicted subsequent child behavior problems at 6 months such that higher levels of perceived social support led to lower levels of child behavior problems, but child behavior did not predict subsequent social support. Research is scant on the potential mechanisms leading from social support to child behavior in the general population. One explanation is that perceived social support influences parenting practices which, in turn, affects child behavior. For instance, Hashima and Amato (56) found that perceived support was negatively associated with punitive parenting practices. Correspondingly, higher levels of parent social support have been associated with increased child praising and less controlling parent behavior (57). Increased social support and a rich social network may expose parents to positive practices or reinforce parenting norms through social pressure (58). The association between parenting practices and perceived support was noted in one study involving parents of children with autism, where perceived social support was correlated with increased perceived limit setting ability, maternal involvement, and satisfaction with parenting (59).

Baseline support was also found to lead to decreased stress at 6 months, though the path from 6-month support to 12-month stress was not significant. This result provides partial support for the hypothesis that social support is a resource that may alleviate parent stress, even when past stress levels and known stressors are controlled (e.g., education level, income, child autism symptoms, adaptive skills). There was no evidence that higher stress levels lead to perceived support. These results are consistent with the single existing study examining this bidirectional relationship longitudinally for mothers of children without neurodevelopmental conditions. Green and Rodgers (60) reported that baseline perceived social support predicted perceived stress 1 year later, but stress did not predict subsequent social support over and above baseline social support. Further, in a longitudinal study involving 283 Canadian mothers of young children with autism, higher perceived social support at baseline was associated with lower levels of subsequent parent stress 2 years later (61), but the opposite effect was not investigated. These findings are consistent with cross-sectional studies.

Limitations

This study has a number of limitations. Participants were recruited through community organizations and a research lab database, and thus parents were likely engaged with autism services or had previously been active in research activities. Parents were mainly well-educated mothers living in suburban or urban locations and nearly all children were born in Canada. Further work with more diverse samples and comprehensive national recruitment strategies is needed as the current study results may not generalize to all parents of children with autism. Second, the data were collected through self-report surveys and it is possible associations among variables are inflated due to shared method variance. We relied on parent report of the autism diagnosis source (e.g., pediatrician, psychologist), diagnosis date, and parent report SCQ scores. Although the SCQ has been found to a valid screener for autism symptoms, in-person diagnostic testing is ideal. Additionally, the current study investigated social support over a 12-month period and future research should study social support over longer periods of time to better understand patterns of change. Furthermore, survey measures used different time periods of reference and this may have influenced the strength of associations. For instance, the measure of stress asked participants to consider the previous week, while the received support measure focused on the previous 4 weeks. Adjusting the time point reference for consistency would be something to consider for future studies. Finally, future studies could examine other dimensions of social support (e.g., social network characteristics, support needs support from specific sources), assess stress within specific contexts (e.g., parenting stress), or consider other social support determinants such as date of autism diagnosis (61), familial interactions from early childhood (30), parenting practices [e.g., (62)], and personal predispositions [e.g., (63)].

Conclusions

After controlling from socioeconomic status, health status and autism symptomology, the stability of perceived social support, parental stress and child behavior challenges were clearly demonstrated. Consistent small to moderate concurrent relationships between perceived social support were and parent stress were found, with minimal evidence for cross-lagged relationships (perceived social support was related to parent stress and behavior problems 6 months later). Examining the concurrent and cross-lagged relationships between perceived social support and child behavior problems, no relationships were found between the 6- and 12-month time point. Given this stability, it is critical that interventions aim to address child behavior problems, stress, and also ways of shifting social support. To our knowledge, within the context of children with autism, there is no existing evidence-based intervention specifically targeting parents' perceptions of their support, though some multi-component programs have incorporated discussions on accessing social support (64–66). Given the presence of both considerable evidence for parenting stress programs (67) and programs to address child behavior problems (68), early access to methods of improving perceptions of social support would be a logical next line of intervention research.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because we are unable to provide access to the dataset related to this work for privacy and consent reasons. Requests to access the datasets should be directed to jonweiss@yorku.ca.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Human Participation Review Committee, York University. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JW was responsible for overall supervision of the project, conceptualization, funding, and final manuscript preparation. SR was responsible for conceptualization, data collection and analysis, and initial manuscript preparation. RP and DF provided guidance for research design, data analysis, conceptualization, and contributed to manuscript preparation. All authors contributed to the article and approved the submitted version.

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Information and Empowerment of Families of Children With Cerebral Palsy in Brazil: The Knowledge Translation Role of Nossa Casa Institute

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Knowledge translation (KT) is gaining attention in the pediatric rehabilitation field. *Nossa Casa Institute* is the first organization in Brazil aiming to foster cerebral palsy (CP) awareness and empower families by discussing reliable information. This study aims to build a network where individuals with CP and their families, researchers, health care professionals, and services can communicate and share experiences. In this article, we describe the experience of planning and conducting an educational and interactive online workshop to foster principles of family-centered service (FCS). We used the action cycle from the Knowledge to Action (KTA) framework to describe and ground the proposed activities. In Module 1, "Challenges and barriers to incorporate family-centered principles," we discussed the historical perspective, main principles, and challenges related to FCS implementation. Module 2, "What is my contribution to the family-centered service?" was aimed to foster strategies to improve the implementation of principles of FCS in the care of children with disabilities. In Module 3, "What can we do together?" the groups presented their ideas and suggestions. This interactive and educational workshop was an opportunity for *Nossa Casa Institute* to disseminate accessible and reliable information regarding FCS and to empower families to participate actively in the rehabilitation process and advocate for the best provision of care for their children. Future actions of *Nossa Casa Institute* include the coordination of a national conference to connect families, individuals with CP, healthcare and rehabilitation professionals, and researchers. There is also a need, and opportunity, for formal evaluation of these KT activities.

Keywords: knowledge translation, family-centered, action cycle, pediatric rehabilitation, cerebral palsy

INTRODUCTION

Knowledge translation (KT) is defined by the Canadian Institutes for Health Research as "a dynamic and iterative process that includes synthesis, dissemination, exchange, and ethically sound application of knowledge" (1). KT aims to diminish the gap between the research literature and its application into practice (2–4). In this process, there should be an articulation between researchers and the end-use stakeholders (i.e., health care professionals, policymakers, and patients) (4–8).

Different models have been proposed to explain the KT process (9). The Knowledge to Action Model (KTA) proposed by Graham et al. (2) has been extensively used in health care studies (10). In the KTA model, KT involves two main elements: knowledge creation and an action cycle (2). Knowledge creation involves knowledge inquiry, synthesis, and the formation of tools and products. The action cycle is composed of activities to allow knowledge application. It encompasses identification of the problem to be addressed, adaptation of the knowledge to the local context, assessments of barriers to knowledge use, selection and implementation of interventions, and monitoring, evaluating, and sustaining knowledge use (2).

Knowledge translation is gaining considerable attention in the rehabilitation field (6–8, 11, 12). The resources and strategies used in KT vary from the provision of single activities (e.g., educational workshops) to multifaceted approaches, with the use of active and multiple tools (e.g., educational workshops and public audit) (6, 11). In pediatric rehabilitation, most initiatives are tailored to overcome the knowledge-to-practice gap, with specific actions directed to health care professionals and services, aimed to promote changes in clinical behavior and to improve care (11, 13–15). Recently, some KT activities with the participation of families have been reported (7, 15, 16).

CanChild Centre for Childhood Disability Research is a university-based health services research program, and an example of an organization pursuing research, education, and KT in pediatric rehabilitation (15–17). The center's interest and engagement with KT have become a major focus of its activities, grounded in the principles of the KTA framework outlined above.

Despite progressive efforts to promote and achieve KT actions in health, many challenges are reported (10, 18–20). Ferraz et al. (18), in a scoping review, synthesized the main challenges. One of the challenges is the lack of cohesion among researchers, populations, and health care policymakers. In this sense, researchers and end-user stakeholders should communicate, so that research questions and methods meet the needs of the community as identified by the community. Second, is the difficulty for health professionals to translate and apply new knowledge. This may be due to the lack of abilities of clinicians or time available to appraise the literature critically and to understand statistical methods. Third, the lack of incentives and supports from health institutions to engage in KT restricts opportunities for continued education of their professionals (18).

Challenges for KT in health are intensified in developing countries (20–22). To the best of our knowledge, there was no structured program in KT in the pediatric rehabilitation field in Brazil before the foundation of *Nossa Casa Institute* in 2016. It is the first online platform to discuss the daily living of individuals with cerebral palsy (CP) and their families, considering the importance to facilitate both communication and implementation of knowledge about CP among all stakeholders in Brazil.

NOSSA CASA INSTITUTE

Nossa Casa Institute is a non-profit organization, funded and started in 2016, that grew out of collaboration among individuals with CP, families, health care professionals, and researchers. The meaning of "*Nossa Casa*" in English is "*Our Home.*" These words were chosen to represent a place for open discussion of information and ideas about CP in a welcoming, friendly, and safe way. This institute aims to build a network where local and international researchers, health care professionals and services, individuals with CP, and their families can communicate, discuss, and share experiences. In 2018, *Nossa Casa Institute* was recognized by the Cerebral Palsy Foundation in the World Cerebral Palsy Day, winning the "Public Awareness" award.

The online tools of *Nossa Casa Institute* include a website, an Instagram account with almost 16,000 followers, a Facebook fan page with 20,800 participants, and 2,500 subscribers on its YouTube account. As a KT Institute, we seek to discuss reliable information regarding issues in the daily lives of people with CP and evidence-based information about assessment and intervention strategies for this population, using plain, accessible user-friendly language. Four groups of activities are conducted by *Nossa Casa Institute:* (1) online interactions, including social media posts and live sessions; (2) development of educational and informative materials, such as videos, awareness campaigns, tutorials, and translated guidelines and worksheets; (3) educational training, workshops and conferences, with national and international experts discussing topics related to CP; and (4) collaboration with researchers to conduct studies aimed at promoting the daily functioning of individuals with CP. **Table 1** shows the main actions developed by *Nossa Casa Institute* since its foundation.

The activities at *Nossa Casa Institute* involve active collaboration among families, individuals with CP, health care professionals, and researchers. Families and individuals with CP participate in online interactions, suggest themes to be discussed, conduct live interactions, and review the content of the shared posts. As for the educational and informative materials, families and individuals with CP collaborate with the conception, illustration, and description of the videos and with content review. Educational training was originally directed to health care professionals and researchers (e.g., General Movement Assessment workshop). In 2019, the International CP Conference was opened to families who showed interest in attending the event. At this conference, one speaker presented his experience as an adult with CP and two mothers of a child with CP spoke. Nowadays, the main efforts of *Nossa Casa Institute* are focused on planning and developing educational training activities with accessible language and active participation of families and individuals with CP. Main representatives

TABLE 1 | Description of main actions and activities developed by *Nossa Casa Institute*, from 2016 to 2021.

Main actions	Activities
Online interactions, including social media posts and live interactions	463 Instagram Posts (@nossacasa.org.br) 40 Instagram lives (@nossacasa.org.br) 57 Youtube videos (Instituto Nossa Casa) Facebook fanpage 1 website (www.nossacasa.org.br)
Development of educational and informative materials regarding individuals with cerebral palsy (CP), such as videos, tutorials, and translated guidelines and worksheets	8 video animations: - "Let's talk about cerebral palsy!", 2,900 views (Available at: https://youtu.be/0qm142gZ3Hc) - "What's cerebral palsy?" 52,400 views (Available at: https://youtu.be/oo4NIPgqLW4) - "F-Words," 10,000 views (Available at: https://youtu.be/xPMzPJwWop8) - "Perinatal stroke," 6,800 views (Available at: https://youtu.be/ml5rEOTUImg) - "Early intervention in hemiparesis," 15,800 views (Available at: https://youtu.be/MpSCPKHVDmw) - "Cerebral Palsy and ICF," 6,900 views (Available at: https://youtu.be/JknMCYaopF8) - "What is evidence-based practice?", 4,700 views (Available at: https://youtu.be/aNck3M5QWqo) - "Family-Centred Service" (to be released in 2021) 2 video tutorials: - "Moving is Power!" (adapted from Go Baby Go) (to be released in 2021) - Low-cost adapters for play (to be released in 2021) - Translation: FCS worksheets from Canchild (to be released in 2021)
Educational training, workshops, and conferences, with national and international experts	- "General Movements Assessment Workshop," conducted by Christa Einspieler, Campinas, Brazil, 2016 - World CP Day Campaign in Brazil (2017, 2018, 2019, 2020) - "Children with CP GMFCS levels IV and V: what should we do?", conducted by Ginny Paleg, Campinas, Brazil, 2018 - International Cerebral Palsy Conference (800 participants, 35 speakers), Campinas, Brazil, 2019 - "Moving is Power!", conducted by Marina Alroldi and Beatriz Vieira, 200 participants, online event - "Family-Centred Workshop," conducted by Marina Brandão, Peter Rosenbaum, Rachel Teplicky, 86 participants, online event, 2020 - "Cerebral Palsy Online Congress,," 1,700 participants, 72 speakers, online event, 2021
Collaboration with researchers and institutions	- CanChild Centre for Childhood Disability, Hamilton, Ontario, Canada - Universidade Federal de Minas Gerais, Belo Horizonte, Brazil

of families and individuals with CP are also collaborators of research initiatives in KT at *Nossa Casa Institute*, including the conception of the studies, data collection, and writing process.

FAMILY-CENTERED WORKSHOP: FROM CHALLENGES TO SHARED SOLUTIONS TO IMPLEMENT FAMILY-CENTERED SERVICES (FCS)

The active participation of families in the rehabilitation of their children with disabilities is widely encouraged in the literature (23–26). To make the best decisions for their children, families should have access to reliable and accessible information and the opportunity to express their preferences, interests, and concerns (23, 24, 27). Although literature on FCS has been available since the 1990s, the implementation of its principles in practice is still challenging. Thus, the proposal of an FCS Workshop with online interactions and educational material in Portuguese (Brazil) aimed to facilitate the incorporation of FCS principles in the pediatric rehabilitation field.

In this "perspective" article, we describe the FCS Workshop to illustrate the role of the *Nossa Casa Institute* in KT activities. We anchored the activities of this workshop in the action cycle of the KTA framework (2) (**Figure 1**).

Identifying the Problem: Preparing for the FCS Workshop

The workshop was moderated by two researchers from CanChild and one researcher from Brazil, who are experienced in developing and implementing FCS. The co-founders of *Nossa Casa Institute* organized two online meetings with the researchers to discuss the workshop format from their previous experience and the relevant content related to the FCS literature. The workshop audience included 16 families of children and adolescents with disabilities, 2 adults with CP, 40 health care professionals, 10 healthcare service coordinators, and 16 academics (i.e., professors, researchers, and students).

The structure of the online workshop included access to pre-recorded educational videos and interactive online sessions. The pre-recorded videos presented preliminary information for the online discussions, as well as providing the participants access to the main literature regarding FCS. Participants were encouraged to access the educational videos prior to each online interaction to create a common starting point for discussions. The online interactions were designed to promote opportunities for all participants actively to build competencies in FCS. Each interaction lasted 1 h on average. The workshop had 3 modules: Module 1: "Challenges and barriers to incorporate family-centered principles"; Module 2: "What is my contribution to family-centered service?" and Module 3: "What can we do together?" The modules were organized so that the participants could share initial thoughts of challenges regarding the FCS principles and then build their own competencies and possibilities to assume family-centered behaviors and attitudes.

Adaptation of the Knowledge to the Local Context: Module 1

The workshop started with an online interaction to present a historical perspective of FCS for children with disabilities

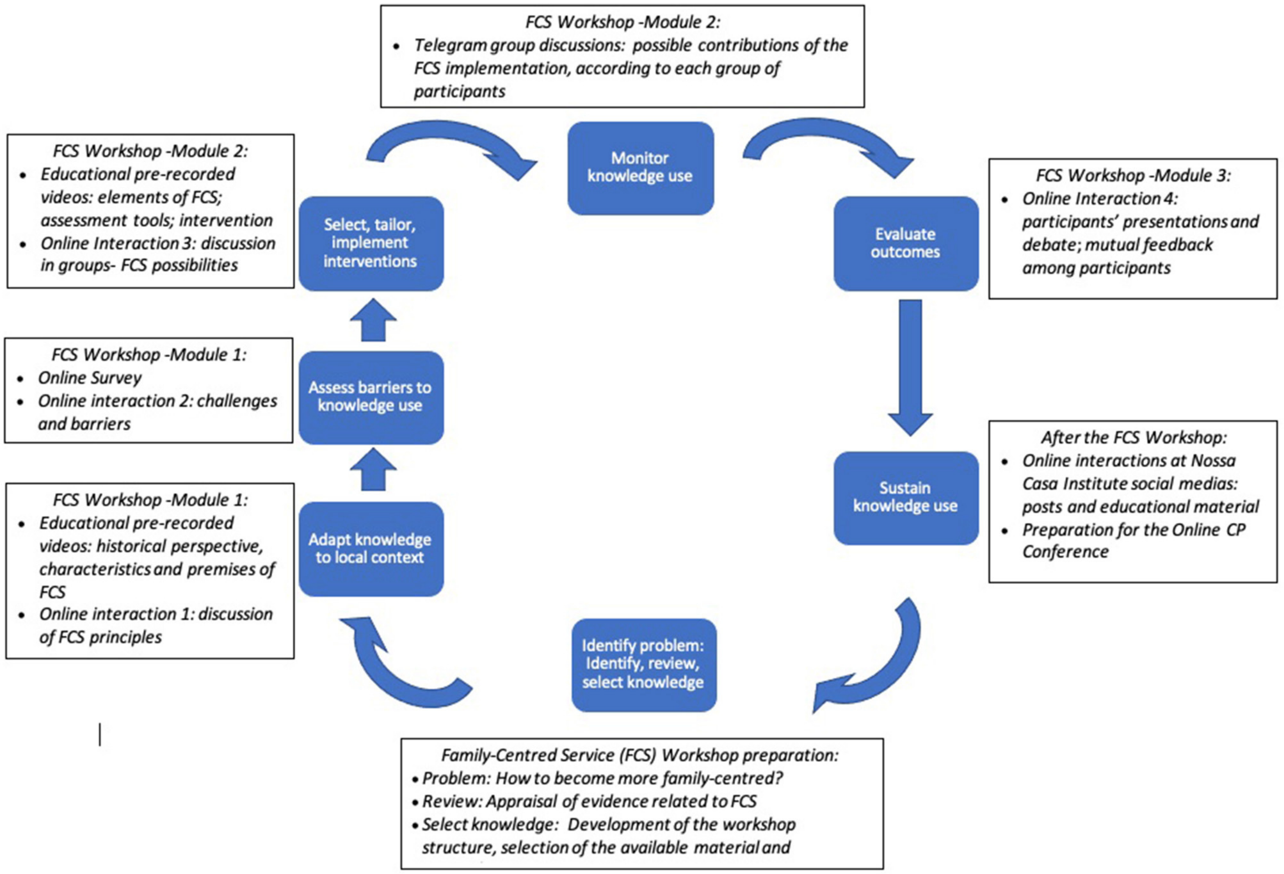

FIGURE 1 | Illustration of the FCS Workshop based on the action cycle from the Knowledge to Action (KTA) model [adapted from Graham et al. (2)].

and debate its main principles. For that purpose, CanChild researchers discussed the main principles and a brief historical perspective of FCS. All participants were encouraged to post their questions and considerations in an online chat to show their opinions and comments. Such interaction facilitated an understanding of the main concerns and expectations of the participants. Three pre-recorded educational videos were available with the main characteristics of FCS.

Assessments of Barriers to Knowledge Use: Module 1

After the first online session, we sent an anonymous online survey to be completed by the participants, built specifically for the workshop. We prepared five surveys following the same structure, but including information specific to the group of the participants (i.e., adults with CP, families of children/adolescents with disabilities, health care professionals, health care coordinators, and academics). In this survey, we asked opinions of the people regarding the main barriers to the implementation of FCS principles in Brazil, and the challenges they faced. We were interested in issues and experience with the services undergone by the children and families. We wanted to hear the experiences of adults with CP in their daily routines and services, and in their interactions with health care professionals, coordinators, and researchers. It was important to learn which FCS principles are already incorporated in the care of their child (families), the care of adults with CP in their daily practices and services with health care professionals, coordinators, and researchers, and which aspects related to FCS they would like to discuss in the workshop.

The information from the online survey of the participants was classified according to challenges related to the behaviors, attitudes, and actions of therapists; behaviors and expectations of families; and the actions of those involved in the format and regulations of services in Brazil. The main concerns reported by the participants were discussed in a second online live interaction to elucidate the possible myths and common misunderstandings underlying FCS.

Selection and Implementation of Interventions: Module 2

Module 2 was designed to discuss strategies to improve the implementation of principles of FCS. We prepared six pre-recorded video lectures in Portuguese (Brazil) based on FCS sheets of CanChild (28): effective communication; building on competencies of families; respect; negotiation; partnership; and decision-making process. The choice for these topics was based on the challenges and barriers that participants reported in the survey. We provided two additional lectures with instrumental information related to FCS implementation: assessment tools to measure FCS outcomes and analysis of family involvement in interventions for children with CP.

The online interactions occurred in four main groups: families and adults with CP, health care professionals, health care coordinators of services, and academics (i.e., professors, undergraduate, and graduate students). In these separate meetings, each group was assigned a specific topic for discussion, with the assistance of a moderator. With families and adults with CP, we asked the participants to discuss their main priorities and needs from rehabilitation services and health care professionals, based on FCS principles. Health care professionals discussed how to improve the family-centered relational components (e.g., behaviors, attitudes, and values) and operational components (i.e., assessment tools and intervention strategies). Health care coordinators discussed ideas to structure their services in light of family-centered characteristics. Academics, including professors, graduate, and undergraduate students, were asked to consider the academic role in implementing FCS.

Monitor Knowledge Use: Module 2

After discussing their main ideas, the participants in each group were asked to prepare a small presentation to be shared in the last meeting. Their interactions had previously been moderated by the Brazilian researcher in Telegram discussion forums.

Evaluate Outcomes: Module 3

In the last online interaction, the groups presented their ideas and suggestions from the separate discussions. All participants were encouraged to present their perspectives to identify effective strategies to promote FCS practices. The first group to present were the "families and adults with CP group," who expressed their needs and their expectations from services and therapists. Parents reinforced their desire to be listened to and trusted by therapists regarding their priorities; they asked for their voices of children to be heard; they believe that their children should be seen as children who need time to play and to have fun; and they reported the desire that therapists should involve not only mothers but also fathers and the extended family in their rehabilitation program. They were also encouraged to express their opinions during the presentations of the other groups. The following three groups (health care professionals, academics, health care providers) presented relational and instrumental strategies they recommended to be adopted in their daily practices. After their presentations, we asked the participants to report their comments and suggestions about their experience at the workshop.

Sustain Knowledge Use: After the FCS Workshop

After the workshop, we invited the participants to join one Telegram group. In this group, they were able to contact each other and share experiences. The information regarding the feedback of the participants in the workshop helped to create online information on the social media channels from *Nossa Casa Institute*. In addition, such information supported the planning and development of an online conference (Online CP Conference: From all to All), aimed at improving the dissemination of knowledge among individuals with CP, families, health care professionals, and researchers. Specific information regarding the online conference and its impact will be reported in future studies.

LESSONS LEARNED

Nossa Casa Institute was conceived to share information about the daily living of individuals with disabilities, with emphasis on CP. It is also creating opportunities for listening and exchanging knowledge through actions on social networks (e.g., interactive video lives, campaigns, and educational workshops) with the participation of all stakeholders. The involvement of families and individuals with disabilities in activities held at *Nossa Casa Institute* has contributed to the greater empowerment of these populations. Furthermore, these actions are enhancing the awareness and education of health care professionals and researchers and reinforcing the value of involving families and individuals with disabilities in research and educational initiatives.

One of the main challenges experienced by the *Nossa Casa Institute* is related to the lack of financial support from the Brazilian government or other agencies; so far, such activities are conducted by volunteering work from its collaborators. Possible subsidies would support future KT activities at *Nossa Casa Institute*. Future studies are planned to explore and evaluate the impact of the proposed actions to support the KT in the pediatric rehabilitation field in Brazil.

DATA AVAILABILITY STATEMENT

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

AUTHOR CONTRIBUTIONS

MB coordinated the project and was involved in the conception, writing, and review of the manuscript. MA, BV, and MM were involved in in the conception, writing, and review of the manuscript. DC and RB are representatives of families and adults with cerebral palsy and they helped in the conception and review of the manuscript. RT and PR were involved in the conception and review of the manuscript. All authors contributed to the article and approved the submitted version.

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Fathers Matter: Enhancing Healthcare Experiences Among Fathers of Children With Developmental Disabilities

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Background: Being a parent of a child with a developmental disability (DD; e. g., cerebral palsy, autism) comes with great challenges and apprehensions. Mothers and fathers of children with DD are experiencing heightened levels of psychological distress, physical health problems, financial difficulties, social isolation, and struggles with respect to traditional parenting roles. In relation to the latter, the involvement of fathers in caregiving in today's society is increasing and is highlighted by its importance and positive contribution to the development of their children. However, fathers of children with DD report feeling excluded and marginalized by healthcare providers (HCPs) when arranging for and getting involved in healthcare services for their children. Currently, there is limited evidence as to what factors influence those experiences. We aimed to explore barriers to and facilitators of positive and empowering healthcare experiences, from the perspectives of fathers of children with DD and HCPs.

Methods: A mixed-method approach, such as quantitative (survey) and qualitative (semi-structured interview) strategies, was used. Participants were fathers of children with DD and HCPs working in childhood disability. Data analysis consisted of using descriptive statistics and an inductive-thematic analysis of emergent themes.

Results: Fathers (*n* = 7) and HCPs (*n* = 13, 6 disciplines) participated. The fathers indicated that while they were moderate to very much satisfied with their interactions with HCPs, they reported that HCPs were only sometimes attentive to them during interactions. Fathers also revealed that positive interactions with HCPs in relation to their children had multiple benefits. Several themes related to barriers and facilitators of optimal interactions and parent–professional relationships emerged. These included session factors (time, attention), personal factors (knowledge of the condition, child and healthcare system, acceptance vs. denial, previous experiences, culture, stereotypes, pre-existing beliefs, stress levels, working schedule), and family dynamics. The participants offered several insights into the different strategies that can be implemented to promote optimal interactions between fathers and HCPs.

Conclusion: We identified several barriers, facilitators, and improvement strategies for optimal interactions and enhanced parent–professional relationships from the perspectives of fathers and HCPs. These can be integrated by existing clinical settings in efforts to enhance current clinical practices and improve child- and parent-related outcomes.

Keywords: health-care experiences, barriers and facilitators, interactions with health-care professionals, clinical practice, family-centered approach, father, children with disabilities

INTRODUCTION

Becoming a parent, in itself, is a stressful life event. Becoming a parent of a child with a disability imposes even greater challenges and concerns, as parents now need to adjust to provide care for a child with emergent and frequently changing healthcare needs [reviewed in (1)]. Both mothers and fathers of children with disabilities are reported to experience heightened levels of stress and psychological distress (2, 3), physical health problems, financial difficulties, social isolation, depression, and conflict with traditional gender roles (4, 5). Moreover, for the last five decades, the amount of time fathers spend with their children has increased dramatically (6). Currently, fathers are acquiring a wide range of roles and responsibilities beyond the traditional ones, where they are becoming more conscious and aware of being models of social and emotional behavior for their children (7). In fact, the involvement of the father in childcare is reported to result in positive socio-emotional, cognitive, and developmental outcomes of their child [reviewed in (8)], as well as in improvements to the emotional well-being and stress levels of the mother (9).

However, despite the increasing involvement of fathers in caregiving, as well as its important and positive contribution to child development, research in the field of childhood-onset disability is still primarily focused on mothers (10–12). For instance, while the applications of family-centered approaches involving parents are on the rise, these are mainly directed toward and largely used by mothers (13). The need to consider fathers, however, and the challenges this can present are increasingly recognized (10). For example, preliminary evidence suggests that fathers of children with disabilities feel excluded and marginalized by healthcare providers (HCPs) when arranging for and getting involved in healthcare services provided for their children (14).

Communication of parents with healthcare professionals in relation to their child with disability reveals a great deal. For example, a pilot study examining a coping intervention for parents of children with disabilities determined that the amount and the quality of the communication with HCPs were a primary predictor of how fathers managed condition-related stress levels (15). Similarly, the quality of parent–professional relationships was hypothesized to influence these interactions (1) and ensure the understanding of the father about the condition of their child (16). Previous efforts have been made to describe the healthcare experiences of fathers of children with (10) and without disabilities (8). Nonetheless, to the best knowledge of the authors, there is limited evidence as to what factors influence the father-healthcare professional interactions, both from the points of view of fathers and healthcare professionals.

With the aim to promote beneficial and helpful interactions and experiences for fathers of children with developmental disabilities, the goal was to explore the barriers to and facilitators of positive interactions and empowering parent-healthcare professional relationships, from the perspectives of fathers of children with developmental disabilities and healthcare professionals (HCPs).

Where,

…*children with developmental disabilities* are children (0–18 years old) with primary motor impairments (e.g., cerebral palsy, muscular dystrophies, spina bifida, spinal muscular atrophies, etc.) and/or with developmental behavioral conditions (e.g., autism spectrum disorders).

…*father* is the male parent in relation to the child (biological, adopted, foster, step).

…*healthcare service experiences* include interactions with HCPs for diagnostic, treatment or medical follow-up purposes related to the disability of the child, in different healthcare settings (e.g., hospitals, rehabilitation centers, community clinics).

…*HCPs* include all-specialty physicians and surgeons, nurses, occupational and physical therapists, speech language pathologists, social workers, special educators, etc.

MATERIALS AND METHODS

Study Design

A mixed-method study design was used, including quantitative (Likert-scale survey) and qualitative (semi-structured interview) methods. In addition, we adopted a patient-oriented research methodology. This was achieved by recruiting two parent advisors to be part of the research team prior to protocol development (DC & FG). They are both fathers of children with disabilities. The parent advisors were involved in the following activities in relation to the project: development of study protocol, development of measurement tools (survey and structure of the interview), data interpretation, and review of ensuing knowledge translation material (presentations, present manuscript).

Study Population

Populations of interest included HCPs and fathers of children with DD. The inclusion criterion for HCPs was to be a licensed HCP, working in the field of childhood-onset developmental disability for ≥6 months, and providing direct assessment and/or intervention for a minimum of 50% of their working time. The exclusion criterion was (1) research or medical assistant personnel (e.g., administrative assistants) who are not directly involved in procedures related to diagnostics and/or treatment and follow-up of children with DD. For fathers of children with DD, the inclusion criteria were (1) male gender and (2) being a father (biological, adopted, step, foster) for ≥6 months of a child with a diagnosed DD (e.g., cerebral palsy). All the participants needed to be fluent in English or in French.

Sample Size Consideration

We aimed to recruit a sufficient number of participants to achieve data saturation. An effort was made to recruit HCPs from different disciplines and fathers of children with different disabilities and various age groups.

Source of Data

The participants were recruited using word-of-mouth/snowball/convenience sampling techniques at the collaborative healthcare services points. These included the Montreal Children's Hospital, Shriners Hospital for Children (Montreal, Quebec, Canada), and the BC Children's Hospital, Sunny Hill Health Centre for Children (Vancouver, BC, Canada). Recruitment was performed via wall-mounted/email/web-posted advertisements. Ethical approval was obtained from all participating clinical sites and informed consent was provided by all the recruited participants prior to their engagement with the study.

Study Procedures

First, the participants were asked to fill out a demographic form and a short Likert-scale based survey that was developed in-house and reviewed by all team members, including patient advisors (**Supplementary Material 1**). For the father-participants, the survey aimed to gather information about their involvement in the healthcare of their child and overall perceptions regarding their experiences and interactions with HCPs in the past. For the HCPs, the survey was designed to gather their perspectives about the involvement of fathers in the healthcare of children and their experiences and interactions with fathers in their clinical practice. Individual interviews were then conducted online using Zoom (Zoom Video Communications, Inc., San Jose, CA, United States). The interviews followed a semi-structured format that was co-developed by the team, including patient-partners (**Supplementary Material 1**). The interviews included introductory statements, open-ended theme-related questions, and interview terminations using closing remarks and a summary of discussed topics/points raised. The interviews ended when no new ideas emerged following the summary/closing remarks statement (i.e., data saturation was reached).

Variables Collected

Demographic variables for HCPs included their discipline and degree, practice clinical setting, amount of experience in the field of childhood disability, age, gender, race/ethnicity, population served, and time spent on continuing education. Demographic variables for fathers of children with disabilities included their age, race/ethnicity, relation to the child (biological, foster, stepparent, adopted), marital status, education level, employment status, and information about the child with a disability (age, condition, rank in the family).

Main variables collected (from the perspectives of both study groups) included (1) the perception of involvement of fathers in healthcare of their child; (2) satisfaction and comfort level in interactions between HCP and fathers of children with disabilities; (3) influential factors (barriers and facilitators) impacting experiences/interactions of fathers of children with disabilities with HCPs; (4) effects of involvement of fathers (from the perspective of HCPs only) and effects of interactions with HCPs (from the perspective of fathers of children with disabilities only) on personal, child-related, and family-related outcomes; and (5) strategies and recommendations to enhance interactions and parent–professional relationships and promote family-centered care.

Data Management and Analysis

Descriptive statistics were used to summarize demographic data. Audio data from the interviews were transcribed verbatim. The NVivo® software (QSR International, Doncaster Australia) was used for data management. Triangulation methods were used for data analysis (17). More specifically, one author (TO) read all the transcripts to gain a general sense of the meaning of the content. The content of the transcripts was then analyzed by generating initial codes for all meaningful ideas emerging from the data using a directed content-based analysis technique (18). Following this, a second coder (research assistant who was not a study participant and did not assist with the interviews in any way) verified the coding grid. Discrepancies were discussed with both raters to explore their meaning and/or relationship to other codes, and a consensus was reached. A final round of analysis was then performed by the first coder to ensure that all relevant statements were coded and that agreement between raters was 100% for all statements.

RESULTS

Demographics of Participants

Seven ($n = 7$) fathers of children with disabilities were recruited. The father-participants were 42.6 ± 8.2 years old, 100% Caucasian, all biological parents to their children, all fulltime workers, four ($n = 4$) were married, and three ($n = 3$) were single. Their education level ranged from a college professional diploma ($n = 3$), Bachelor ($n = 3$) to Masters ($n = 1$). Their children with disability (male: $n = 4$; female: $n = 3$) were on average 9.4 ± 5.3 years old (range: 18 months—15 years) with the following

TABLE 1A | Response frequencies of father-participants: involvement in healthcare and satisfaction/comfort level of child in interactions with HCPs.

	Not all all **Never** ***n* (%)**	**Slightly** **Sometimes** ***n* (%)**	**Moderately** **Often** ***n* (%)**	**Very much** **All the time** ***n* (%)**
I am involved in setting up/arranging/organizing health-care services for my child.	2 (28.5)		2 (28.5)	3 (42.8)
I am involved in taking my child to his/her health-care services appointment(s) and/or emergency visit(s).		2 (28.5)	2 (28.5)	3 (42.8)
I am present (with my spouse/partner, if applicable) when my child is receiving health-care services.		1 (14.2)	4 (57.1)	2 (28.5)
I advocate for my child's health-services.	1 (14.2)	2 (28.5)		4 (57.1)
I am taking part in applying treatment recommendations that my child receives			2 (28.5)	5 (71.4)
Overall, I am satisfied with the interactions/communication I had with HCPs in the past.		2 (28.5)	2 (28.5)	3 (42.8)
HCPs are generally attentive to me.		4 (57.1)	1 (14.2)	2 (28.5)
I feel generally comfortable with the HCPs.		1 (14.2)	1 (14.2)	5 (71.4)
I feel generally understood by the HCPs.		1 (14.2)	3 (42.8)	3 (42.8)
I feel generally supported by HCPs.		2 (28.5)	3 (42.8)	2 (28.5)
I feel that generally I formed and continue to have good relationships with the HCPs.			3 (42.8)	4 (57.1)
I had positive/good experiences/helpful interactions with HCPs.			1 (14.2)	6 (85.7)
I had negative/bad experiences/not helpful interactions with HCPs	4 (57.1)	1 (14.2)	2 (28.5)	

Response frequency (%)	**Color-code**
0 to <25	░
25 to <50	▒
50 to <75	▓
75–100	█

conditions: epilepsy, anxiety, cognitive and language delay, trisomy 21, cerebral palsy, autism spectrum disorder, attention deficit and hyperactivity disorder, developmental coordination disorder, behavioral challenges, and globoid cell leukodystrophy.

Thirteen ($n = 13$) HCPs were recruited. HCPs (female: $n = 10$; male: $n = 3$) were 37.8 ± 13 years old. Degrees obtained included Bachelor ($n = 1$), Masters ($n = 8$), Medical doctor ($n = 1$), and PhD ($n = 2$), with diplomas obtained anywhere from 1982 to 2017. The HCPs were from the following disciplines: Occupational Therapy ($n = 6$) Speech Language Pathology ($n = 2$); Neuropsychology ($n = 2$), Pediatrics (Medical Doctor, $n = 1$), Social Work ($n = 1$), and Nursing ($n = 1$). Most of them were working fulltime ($n = 10$) in the field of childhood disability for an average of 11.5 ± 9.7 years. Fields of practice included general neurodevelopment and autism spectrum disorder ($n = 7$), orthopedics ($n = 2$), neurotrauma ($n = 2$), intellectual disability ($n = 1$), and complex care ($n = 1$). Clinical settings included university teaching hospitals ($n = 5$), rehabilitation centers ($n = 3$), community or private clinics ($n = 3$); and a mix of clinical settings ($n = 2$). On average, the recruited HCPs spent 10.2 ± 12.3 h per month on continuing education.

Survey Responses: Involvement of Fathers in Healthcare and Satisfaction/Comfort Level of Child in Interactions With HCPs

Tables 1A,B outline response frequencies to the initial survey regarding the involvement of fathers in the healthcare of their child, and satisfaction/comfort level in interactions from the perspectives of both groups.

Father-participants reported that 57.1 to 85.7% of them were "Moderately (Often)" to "Very much (All the time)" involved in the healthcare of their child, such as organizing services, taking the child to their appointments and medical visits, being present during medical visits, and advocating for the healthcare of their child. All of them (100%) reported that, at all times, they are taking part in applying treatment recommendations that their child receives. In terms of their satisfaction and comfort level in interactions with HCPs, father-participants reported that 71.3 to 85.7% of them are "Moderately" to "Very much" satisfied with the interactions they have had with HCPs in the past, where they conveyed feeling comfortable, understood, and supported by HCPs. However, 57.1% reported that HCPs are only "Sometimes" attentive to them during interactions. All of them reported that they were able to form a good relationship with HCPs and had a positive/good experience/helpful interaction(s). Only 28.5% of them reported having frequent negative experiences in interacting with HCPs in relation to the care of their child.

On the other hand, 92.3 to 100% of the HCPs reported that fathers are only "Slightly (Sometimes)" to "Moderately (Often)" involved in advocating for the healthcare needs of their child, implementing and applying the recommendation/treatment regiments, getting involved in setting up healthcare services for their child, taking the child to medical visits, and being present during appointments. On average, 83.1% of the HCPs reported that they are satisfied with the interactions they have had with

TABLE 1B | Response frequencies of HCPs: satisfaction/comfort level in interactions with fathers and involvement of father in healthcare of child.

	Not all all Never *n* (%)	Slightly Sometimes *n* (%)	Moderately Often *n* (%)	Very much All the time *n* (%)
Overall, I am satisfied with the interactions/communication I had with fathers of children with disabilities in the past.		3 (23.0)	6 (46.1)	4 (30.7
Fathers of children with disabilities are generally attentive with me during interactions.		3 (23.0)	7 (53.8)	3 (23.0)
I feel comfortable interacting with fathers of children with disabilities.		1 (7.6)	2 (15.3)	10 (76.9)
I feel that fathers of children with disability understand me.		3 (23.0)	5 (38.4)	5 (38.4)
I feel that fathers of children with disabilities feel supported by me.		4 (30.7)	4 (30.7)	5 (38.4)
I feel that I form/continue a good relationship with fathers of children with disabilities.		1 (7.6)	5 (38.4)	7 (53.8)
The extent to which fathers…				
…advocate for their child.		4 (30.7)	9 (69.2)	
…implement and apply your recommendations/treatment regiments you are providing them with.		6 (46.1)	7 (53.8)	
…are involved in setting up/arranging/organizing health-care services for their child.		9 (69.2)	3 (23.0)	1 (7.6)
…are involved in taking their child to his/her health-care services appointment(s) or emergency visit(s).		6 (46.1)	7 (53.8)	
…are present when their child is receiving health-care services (alone or with spouse/partner).		6 (46.1)	6 (46.1)	1 (7.6)

Response frequency (%)	**Color-code**
0 to <25%	░
25 to <50	▒
50 to <75	▓
75–100	█

fathers of children with disabilities. HCPs reported that fathers are generally attentive to them and understand them, and that they (the HCPs) were comfortable in those interactions and were able to form good relationships. However, a lower percentage of the HCPs (69.2%) conveyed that fathers of children with a disability felt "Moderately" (30.7%) to "Very much" (38.4%) supported by them.

Semi-structured Interviews

The initial agreement between the two independent coders of the semi-structured interviews was high at 96.6%. The few discrepancies in coding were resolved through discussion to reach a 100% consensus.

Four main themes emerged from the semi-structured interviews:

1. Impacts of the interactions between fathers of children with disabilities and HCPs (from the perspectives of the father-participants).
2. Impacts of fathers in the healthcare of their child with a disability (from the perspectives of the HCPs).
3. Barriers and facilitators to optimal and empowering interactions between fathers of children with disabilities and HCPs.
4. Solutions to optimize: (a) interactions between fathers of children with disabilities and HCPs and (b) involvement of fathers in the healthcare of their child.

Those four themes and their respective subthemes are described below, along with the most salient utterances from the study participants.

Impacts of Interactions (From the Perspectives of Father-Participants)

Father participants (FP) reported several impacts of positive ($n = 6$ subthemes, $n = 14$ utterances) and negative ($n = 4$ subthemes, $n = 7$ utterances) interactions with HCPs in relation to their child. Impacts of positive interactions included "Gaining a better perception and awareness of child's needs, challenges and strengths" ($n = 7$ utterances):

> *FP3: "I think generally when I walk away from [positive interactions with HCPs], I am being reminded about [my child's] issues and that tends to have a positive effect generally on how I interact with [my child]."*

The participants also reported that positive interactions allowed them to "Focus on what is important" ($n = 2$ utterances) and "Bringing the family closer together" ($n = 1$ utterance):

> *FP3: "Dealing with an issue with [my child] is something that is a scary trying thing on the family, and I think things like that tend to bring us closer together, and [my wife] and I always bond in a positive way over you know what's going on with [our child] and it helps you set aside the sort of small issues that you are having*

> *during the day, you know the small stuff suddenly doesn't seem so important. It tends to bring us closer together, I think".*

Improvements to their "Role as a father" ($n = 2$ utterances), "Relationship with their child" ($n = 1$ utterance) and their "Overall mood" ($n = 1$ utterance) were also conveyed:

> *FP3: "[Positive interactions with HCPs] tend to make me act a little more [with my child]. It kind of brings out the best in me as a father."*
> *FP3: "Those interactions you do have with HCP, they are very significant, and I think that, I'm sure those professionals they deal with hundreds of people a week, but for us as parents, it's a big deal, so if you have a good interaction and somebody is kind and considerate it definitely can influence your overall mood [...]."*

Impacts of negative interactions with HCPs included fathers becoming more "Protective" ($n = 3$ utterances), "Assertive" ($n = 2$ utterances) and "Vigilant" ($n = 1$ utterance):

> *FP2: "[Previous negative experiences with HCPs] resulted in the fact that I was checking everything, verifying if there were any errors. When there was even a small error or inconsistency, I would jump on the opportunity to complain and report it. I think this contributed to us receiving the services we needed [for my child]."*

One participant also reported that negative interactions with HCPs had the potential to "Affect subsequent relations with their partner and/or their child" ($n = 1$ utterance):

> *FP3: "[...] if you have a negative interaction, it can definitely stick with you and can kind of come out with the wife or the kids."*

Impacts of Fathers in the Healthcare of Their Child With Disability (From the Perspectives of HCPs)

The HCPs reported that the involvement of fathers in the healthcare of their child with a disability had numerous positive impacts ($n = 6$ subthemes, $n = 30$ utterances). The most commonly described effect is that fathers have the potential to "Contribute to the child's development, recovery and well-being" when they are involved and engaged in the healthcare of the child ($n = 11$ utterances):

> *HCP05: "I think [fathers] have a significant role [...]. We have limited amount of time to facilitate recovery or adaptation with compensatory measures, it only moves the needle so far. If we can extend that with dad's support, so if they are involved if they are supportive, that facilitates I think in general quicker improvement, quicker recovery, quicker restoration of function and independence. And when I've seen it, it's been amazing."*

Second, the HCPs convey that when fathers are involved, this leads to a "Relief in burden of care for the mother and promotes mother's well-being" ($n = 9$ utterances).

> *HCP03: "[Father's contribution to mother's wellbeing] is huge, it's the biggest role that the father can have, because [he is] the immediate partner, and the immediate support system that the mother has, so it would be a huge impact."*

Several participants mentioned that fathers can promote "Therapy through play and fun" ($n = 5$ utterances) and "Can be very innovative" ($n = 2$ utterances):

> *HCP13: "One of my favorite dad moments was working with a child who was not yet a word user. So, I was having dad say the word 'up' and then toss the kid up in the air, gently, with lots of care and love, and that child did eventually say the word 'up.' Me and the dad thought that might have been the child's first word after maybe about half an hour of that rough and tumble physical play. This was a nice match of a style of play that the dad was comfortable with. 'Up' is a great first word because it's easy and meaningful."*
> *HCP01: "Dads' ideas are really great and dismissing them or not knowing how they are contributing - I think we are losing a big piece of information. Because you see [...] the strategies they think of might be something totally out of your ball game, so they are giving a completely different perspective that could be excellent, so by disregarding them you are missing that."*

The involvement of fathers can also lead to more "Compliance in therapy" ($n = 2$ utterances) and enhanced "Family dynamics and overall functioning" ($n = 1$ utterance).

> *HCP10: "The thought that I had would be the kid who gets really frustrated when either they don't understand what is going on or they can't communicate or both [...] if the dad explains to the other kids and to the grandparents 'here's how we deal with little Johnny,' and if little Johnny feels supported and understood as much as possible, I think that probably has a positive effect on the child's behavior; therefore, the whole family's functioning. It would just sort of reduce the frustration for the child [and will make] everyone a little happier."*

Barriers and Facilitators to Optimal and Empowering Interactions Between Fathers of Children With Disabilities and HCPs

Tables 2A,**B** outline the emerging subthemes related to barriers and facilitators to optimal and empowering interactions between fathers and HCPs from the perspectives of both study groups. Overall, 147 ($n = 147$) utterances were classified in barriers ($n = 87$ utterances; HCP-reported $n = 68$; father-participants-reported $n = 19$) and facilitators ($n = 60$; HCP-reported $n = 41$; father-participants-reported $n = 19$).

The most commonly reported barrier to optimal and empowering interactions between fathers and HCPs ($n = 34$ utterances, accounting for 39.1% of all barriers) was found to be associated with *Father-related factors*. Predominantly, accounting for 76.5% of barriers, these factors included fathers who were uninvolved or disengaged; rigid working schedule and high stress level of fathers; certain personality traits (e.g., being demanding, dismissive, or too imposing); and being in denial with regard to condition and challenges of their child:

> *HCP01: "There are two general types of dads that are less helpful: one is the one that is uninvolved – that's the one that even if they are present, they might be on their phone the whole time not paying attention. You kind of feel like you are bothering them if you have to ask them a question or if you want to point something out to them that their child is doing... those dads are not that helpful. The*

TABLE 2A | Barriers to optimal and empowering interactions and parent–professional relationships between fathers of children with disabilities and HCPs.

Theme ***n*** **= number of utterances % of Barriers**	**Subtheme**	**Description**	**Father's utterances** ***n*** **(% of total)**	**HCPs' utterances** ***n*** **(% of total)**	**Total utterances** ***n*** **(% of Theme)**
Factors related to fathers *N* = 34 39.1%	Uninvolved or disengaged	Fathers who are uninvolved and/or disengaged in the healthcare of their child (e.g., during medical visits)	0 (0)	7 (100)	7 (20.5)
	Rigid working schedule and stress level	Fathers' working schedule and stress levels that limit the possibility of interactions and involvement in the healthcare of their child.	2 (28.5)	5 (71.4)	7 (20.5)
	Unhelpful personality traits	Fathers' personality traits impeding communication and parent-professional relationship building (e.g., too demanding, dismissive, imposing).	0 (0)	6 (100)	6 (17.6)
	Denial	Fathers who are in denial about their child's challenges or who have difficulty accepting the situation.	1 (16.7)	5 (83.3)	6 (17.6)
	Lack of understanding	Fathers who lack understanding or knowledge about their child and/or about the condition of their child.	1 (25.0)	3 (75.0)	4 (11.7)
	Previous experiences	Fathers with previous negative experiences with HCPs and/or the healthcare system and who are "carrying the baggage" into a new conversation/interaction.	0 (0)	2 (100)	2 (5.8)
	Difficulty opening up	Fathers who have difficulty opening up about certain issues and/or share their feelings and concerns for fear of being perceived "weak."	0 (0)	1 (100)	1 (2.9)
	Difficulty understanding HCPs	Fathers who have difficulty understanding HCPs for various reasons (e.g., communication barrier, low education level)	0 (0)	1 (100)	1 (2.9)
		Total	**4 (11.7)**	**30 (88.2)**	**34 (100)**
Cultural beliefs *N* = 18 20.7%	Father's role	The cultural beliefs about father's role in the family and the healthcare of the child.	2 (28.5)	5 (71.4)	7 (38.8)
	HCP who are female	Father's cultural beliefs toward HCPs who are female.	0 (0)	4 (100)	4 (22.2)
	Culture-general	Ethnicity or general cultural factors that can affect interactions.	0 (0)	3 (100)	3 (16.6)
	Disability	Fathers' cultural beliefs about their child's disability.	0 (0)	2 (100)	2 (11.1)
	HCP discipline	Fathers' cultural beliefs about HCP discipline (e.g., physicians vs. therapists).	0 (0)	1 (100)	1 (5.5)
	HCP age/experience level	Fathers' cultural beliefs about HCP of young age and/or possible low level of experience in childhood disability.	0 (0)	1 (100)	1 (5.5)
		Total	**2 (11.1)**	**16 (88.8)**	**18 (100)**
Stereotypical thinking or assumptions *N* = 9 10.3%	**-**	Refers to preconceived or stereotypical beliefs (e.g., "father is present at the medical visit because of lack of trust or unsatisfaction with services").	3 (33.3)	6 (66.7)	9 (100)
		Total	**3 (33.3)**	**6 (66.7)**	**9 (100)**
Family dynamics *N* = 8 9.1%	**-**	Refers to various limiting family dynamics (e.g., overpowering partner).	0 (0)	8 (100)	8 (100)
		Total	**0 (0)**	**8 (100)**	**8 (100)**
Factors related to the healthcare and child-care systems *N* = 7 8.0%	Accessibility to information, services, or contacts	Challenges related to accessing information, services or contact within the electronic record system/scheduling system.	3 (60.0)	2 (40.0)	5 (71.4)
	Delays in appointments - long waiting times	Long waiting lists for medical appointments.	1 (100)	0 (0)	1 (14.2)
	Maternity vs. paternity leaves	Refers to a longer vs. shorter maternity vs. paternity leave, possibly affecting father's involvement in child's healthcare, general care, knowledge about child and condition.	0 (0)	1 (100)	1 (14.2)

(Continued)

TABLE 2A | Continued

Theme *n* = number of utterances % of Barriers	Subtheme	Description	Father's utterances *n* (% of total)	HCPs' utterances *n* (% of total)	Total utterances *n* (% of Theme)
		Total	**4 (57.1)**	**3 (42.8)**	**7 (100)**
Factors related to HCPs *N* = 6 6.8%	Focus on negative aspects	HCP focusing on negative aspects, disability.	2 (100)	0 (0)	2 (33.3)
	Lack of attention to fathers	HCP not directing attention specifically to fathers in interactions.	1 (50.0)	1 (50.0)	2 (33.3)
	Lack of knowledge about cultural factors and how to bridge them	HCP who lacks awareness about certain cultural factors that can be at play and how to bridge them during interactions.	0 (0)	1 (100)	1 (16.7)
	Early career, confidence level	HCP who is early in their career and have a low confidence level when interacting with fathers.	0 (0)	1 (100)	1 (16.7)
		Total	**3 (50.0)**	**3 (50.0)**	**6(100)**
Time limitation *N* = 5 5.7%	-	Refers to time limitations during interactions (e.g., feeling of being rushed through medical visit or limited appointment time because of ongoing case load of the HCP).	3 (60.0)	2 (40.0)	5 (100)
		Total	**3 (60.0)**	**2 (40.0)**	**5 (100)**
		Grand Total	**19 (21.8)**	**68 (78.2)**	**87 (100)**

> *second type of dad that is not that helpful is the authoritative dad who thinks that he knows best because he is the parent. I've had a dad pull me out of a room before I see a child and tell me: 'Tell my kid he can't drive.' whereas that might not be the situation, but he thinks his son cannot drive and therefore I should be conveying the same message. So those are the two general types of dads who are a little less helpful in situations. And also, it puts a strain on the therapeutic relationship because if you aren't going to carry through for ethical reasons or professional reasons you are deemed a bad therapist. And then there is of course stress and work if dads are very-very busy, they may not feel the importance to put down their phone to listen to what we are saying [...]. So, they are stressed and thinking of many-many things, they might miss appointments, and you as the health care professional have the impression that they are not involved but it's not necessarily true because they might be very involved at home it's just that they can't miss work. [...] Stress and work can really affect the way a dad might come across. Maybe they are stressed about something external, and they are taking it out on whoever is in front of them. You don't know what has happened while they have been waiting in the waiting room."*
>
> *FP06: "Yeah, I feel like my ex-wife was accepting my kid like she was. She was more supportive than I was. I was engaged but I was demanding. She was engaged, but she was supportive. I was trying to push [my child] and my ex-wife was trying to support her. When you come in front of a professional, you can't keep pushing; you have to start accepting so you can help. It took me time. [...] I heard quite a bit of that, of people telling me that's not a real issue. So sometimes, the first reaction you get from men is not a very supportive one, so I guess when you go meet a professional if you don't believe in what you are doing and you just doubt the reality of that, it's going to be difficult."*

The remainder of *Factors related to fathers* included lack of understanding or knowledge of fathers regarding the condition of their child; their previous negative experiences with HCPs and/or the healthcare system; difficulty opening about issues and feelings with the HCP; and difficulty understanding the HCP:

> *FP03: "[...] In my experience, [my wife] is always the one with the information and the background knowledge and previous experience with these people and therefore is getting a lot more value out of the meetings. For me, if I don't have the base knowledge of what we are talking about then I'm not going to get as much out of it."*
>
> *HCP09: "So what I find most difficult is when you see that something is bothering [dads] and then they are just not talking. [...] It is hard to get them to confide because they do not do that necessarily, that's not in their nature really to share their feelings, so, but you can actually see that something is wrong and they're just not talking about it. So, it's hard to tiptoe around the issue where you know something is not right, but then, that's another thing, a mom would just come out and tell you what's wrong and what's bothering her and then you can talk about it and get it out and try to find solutions, but with dads it's much more difficult to do that."*

Cultural beliefs, such as those about role of a father, disability, and HCP discipline/gender/age-experience level, accounted for 20.7% of all barriers:

> *HCP10: "I see a lot more different cultures, and over the years it is often, not always, the mother who is the one who describes. Like the dad sort of drove her there, but he doesn't answer a lot of my questions, often isn't that worried, and sometimes I think that is a cultural thing that it's this is the moms job, the mom raises the kids and I make the money or I'm the provider or am the patriarch, but that's their culture for the father not to be as involved and also not that worried about developmental disabilities."*

TABLE 2B | Facilitators to optimal and empowering interactions and parent–professional relationships between fathers of children with disabilities and HCPs.

Theme *n* = number of utterances % of Facilitators	Subtheme	Description	Father's utterances *n* (% of total)	HCPs' utterances *n* (% of total)	Total utterances *n* (% of Theme)
Factors related to fathers *N* = 29 48.3%	Engagement	Father who is engaged and involved in child's care and healthcare.	0 (0)	5 (100)	5 (17.2)
	Ability to see the overall picture and practicality	Father who can see the overall picture and be practical with solutions, innovative.	0 (0)	5 (100)	5 (17.2)
	Acceptance	Father who accepts the child's conditions and challenges.	1 (25.0)	3 (75.0)	4 (13.7)
	Knowledge of the child's condition, medical history	Father who had a good understanding and awareness of the child's condition, medical history and child's needs.	0 (0)	4 (100)	4 (13.7)
	Personality	Helpful personality traits (e.g., advocacy, perseverance, authority).	2 (66.7)	1 (33.3)	3 (10.3)
	Responsibilities sharing	Father who is sharing responsibilities with partner (cultural shift in traditionally set roles).	0 (0)	2 (100)	2 (6.8)
	Ability to prioritize	Father who is able to prioritize needs.	0 (0)	2 (100)	2 (6.8)
	Openness to learn	Father who is open to learn.	1 (50.0)	1 (50.0)	2 (6.8)
	Ability share feelings	Father who is able to open up and share feelings, talk about issues.	0 (0)	1 (100)	1 (3.4)
	Working conditions	Employer who understands father's situation and is flexible, allowing father to be involved in child's healthcare needs.	1 (100)	0 (0)	1 (3.4)
		Total	**5 (17.2)**	**24 (82.7)**	**29 (100)**
Factors related to HCPs *N* = 23 38.3%	Providing explanations	HCP who is providing explanations during interactions with fathers of children with disabilities.	4 (100)	0 (0)	4 (16.6)
	Taking their time in the interactions	HCP who is taking their time during interactions with fathers and do not "rush the appointment."	1 (25.0)	3 (75.0)	4 (16.6)
	Using family-centered care approach	HCP who is using a family-centered approach, where the needs of all family members are heard and considered, where both parents are treated equally.	3 (75.0)	1 (25.0)	4 (16.6)
	Level of experience (novice vs. seasoned)	HCP with more years of experience in the field of childhood disability.	0 (0)	3 (100)	3 (12.5)
	Having the intent to involve fathers	HCP with intent to involve fathers of children with disability into assessment, treatment plan and/or follow-up.	0 (0)	2 (100)	2 (8.3)
	Reporting improvements, focus on positive aspects	HCP who is not solely focusing on disability and challenges, but who also report on positive aspects and improvements.	1 (50.0)	1 (50.0)	2 (8.3)
	Advocating for family's needs	HCP who is acting as an advocate for the family and the child.	2 (100)	0 (0)	2 (8.3)
	Grabbing the opportunity when father shows interest	HCP who can detect father's interest and take the opportunity to involve them as soon as possible.	0 (0)	1 (100)	1 (4.1)
	Engaging fathers	HCP who is engaging fathers as much as possible.	1 (100)	0 (0)	1 (4.1)
	Having a positive father role model	HCP who themselves have had a positive father role model in their life.	1 (100)	0 (0)	1 (4.1)
		Total	**13 (54.2)**	**11 (45.8)**	**24 (100)**
Communication strategies *N* = 4 6.6%	Everyday examples	Using examples from everyday life to describe complex situations.	0 (0)	1 (100)	1 (25.0)
	Visual supports	Using visual supports in interactions that require deep understanding and lots of explanation.	0 (0)	1 (100)	1 (25.0)
	Humor	Using humor during interactions, where appropriate.	0 (0)	1 (100)	1 (25.0)
	Concreteness	Emphasizing how a father can help *via* concrete and clear suggestions and explanations why it is helpful.	0 (0)	1 (100)	1 (25.0)
		Total	**0 (0)**	**4 (100)**	**4 (100)**
Long lasting relationship with the same HCP *N* = 1 1.6%	-	Refers to having a long-lasting parent-professional relationship with the same HCP.	1 (100)	0 (0)	1(100)

(Continued)

TABLE 2B | Continued

Theme *n* = number of utterances % of Facilitators	Subtheme	Description	Father's utterances *n* (% of total)	HCPs' utterances *n* (% of total)	Total utterances *n* (% of Theme)
		Total	**1 (100)**	**0 (0)**	**1 (100)**
Follow-up *N* = 1 1.6%	-	Refers to following up with fathers if they are absent from the medical visit in relation to their child's care.	0 (0)	1 (100)	1 (100)
		Total	**0 (0)**	**1 (100)**	**1 (100)**
Accommodating father's schedule *N* = 1 1.6%	-	Refers to accommodating father's schedule in the attempt to have them present during healthcare visits.	0 (0)	1 (100)	1 (100)
		Total	**0 (0)**	**1 (100)**	**1 (100)**
		Grand Total	**19 (31.7)**	**41 (68.3)**	**60 (100)**

Stereotypical thinking or assumptions; *Family dynamics* (e.g., overpowering partner); *Factors related to the healthcare and childcare system* (e.g., wait times, accessibility to services); and *Time limitation* (e.g., feeling rushed through the appointments) were also reported as barriers:

> *FP02: "Every time the physiotherapist sees me [the father], she has this face: 'Oh okay, it's the father…' as if I were there to be mean, to see how you do your job. I have the impression that it looks like when they see the father [at the medical visit], they think it is because there is something wrong."*
> *FP03: "One thing that always bothers me is when I sense that there is a rush on the meeting, if there is a rush on the interaction we are having, and if there's not enough thought going into a suggestion. You have just met my kid, you are telling him this thing right here, and it is like hang on, there can be an impact there on me, where I feel like, you know you are more apt to follow someone's advice or hear what they have to say if you believe that they know what you are dealing with, they know your particularities. Going to any HCP where they are trying to rush you out of the office it's not a good situation for any positive interaction."*

Lastly, certain *Factors related to HCPs* also emerged as obstacles and included: HCPs who are focusing on negative aspects and on disability; those lacking attention to fathers in their interactions; the lack of knowledge about cultural factors and how to bridge them; and being early in their career (e.g., low confidence level in interacting with fathers):

> *FP03: "I feel like it's just important to remind [HCPs] that we as parents can really use a dose of positivity, and it feels good when they, I think starting out on a foot of positivity has been great and too often people jump straight into 'Okay, here's what I'm seeing that's wrong with your son.' […] it's all about what he's sort of failing at, really, and that can be really stressful actually."*
> *HCP13: "I've seen professionals that are great at coaching moms […] but in other situations, just totally stone-wall dad. What are you doing? He came to the appointment, like you might hook him!"*
> *HCP13: "I have a child on my list right now who, they speak [another language]. During one of my feeding visits, dad is very polite and very quiet, just kind of smiles and nods. And in one of my feeding visits, I said 'I like seeing you, I like seeing you here, I'd love to see you, you know for communication visits, you are so invited, you could come if you wanted to, we'd have fun together,' and then he came to some subsequently which really made [me happy]. But I do not always know how to build that bridge, especially if I do not meet dad in the clinic area in the first place. I don't always know how to build that bridge."*
> *HCP10: "When I was new, I was probably intimidated talking to anyone, I felt like an imposter, like I didn't know what I was doing."*

In terms of facilitators, *Factors related to fathers* were reported 48.3% of the time and included: engagement of father; ability to see the overall picture and practicality; acceptance of the condition of child, strengths and weaknesses; knowledge of the condition of the child; positive personality traits (e.g., advocacy, perseverance); sharing of responsibilities; ability to prioritize; openness to learn; ability to share feelings; and flexible working conditions:

> *HCP10: "The [experiences and interactions] that are positive are [with] the involved dads. They are the ones who are asking me questions who seem to know their child quiet well, they are aware of their child's strengths and weakness."*
> *HCP11: "So the father would come to some appointments and the positive interaction was [that] he was always [sharing] with me how he felt about the kid. He was always very happy when he came back and he had put into place the recommendations I had given him."*
> *HCP01: "[Fathers] still care for their child very-very much, but they aren't caught up in their insecurities, and their guilt, and their anxieties. They are able to take a step back and really see objectivity what is important in this moment for their child."*
> *HCP12: "…Father's openness and willingness to receive information will have a play on how comfortable I am too. So, with the fathers who are really interested and want to know why we are doing certain activities, [the interaction] is really not a problem."*

Several *Factors related to HCPs* emerged as enablers and included: providing explanations; taking their time in the interactions; using a family-centered approach; level of experience; intent to involve fathers; reporting improvements and focus on positive aspects; advocating for needs of the family; grabbing on the opportunity when the father shows interest; engaging fathers; and having a positive father role model in their lives. Seventy percent (70%) of all the utterances of the father-participants in the theme "Facilitators" were found to be in this category (i.e., *Factors related to HCPs*):

> *FP04: "I would more say was the one where we did the occupational therapy where the HCP was actively engaged with the parents, they showed us the techniques to do at home in order to help Jackson when he is at home not just at the therapy sessions, so for me they were engaged with us."*
> *FP03: "[One facilitator to optimal interactions is to] try as best as you can to give time to the interaction, to provide knowledge in the meeting itself for when it's needed, like if, I mean it's definitely not on them to educate us every time, but it's helpful and I appreciate it."*
> *FP02: "[In the best interactions], we [father and mother] were considered both as parents of equal value, and not 'the mother and the father who is on the side.'"*

Further, *Communication strategies* were reported as positive (100% by HCPs) and included: the use of everyday examples; visual supports; and humor; and concreteness, where the HCP can emphasize how a father can help using clear suggestions and explanations as to why it is beneficial.

> *HCP09: "So a lot of the good interaction that I find is when we do teach or we're trying to solve issues, they are very engaged in that. So, to give them concrete explanations, to give them concrete things to do, like, that's what they like, and that's what they're engaged into."*
> *HCP07: "I use humor a lot with fathers, they get on well.. Sometimes when we use a little bit with humor, it creates good interactions."*

The remaining facilitators included having a *Long-lasting relationship with the same HCP*; *Following-up with fathers* when they are absent from the medical visit; and *Accommodating father's schedule* in attempt to have them present during healthcare visits.

Solutions to Optimize Interactions Between Fathers of Children With Disabilities and HCPs and Involvement of Fathers in the Healthcare of Their Child

Seventy ($n = 70$) utterances ($n = 18$, 25.7% from the father-participants; $n = 52$, 74.3% from the HCPs) were categorized into the theme of *Solutions to optimize interactions between fathers of children with disabilities and HCPs and overall father's involvement in the healthcare of their child*. **Table 3** outlines the reported solutions. Most solutions (65.7%) were proposed directly for HCPs and included: *Changes to communication strategies*; *assessment*, *treatment and follow-up*; *Appointments and scheduling*; *Fair consideration of both parent figures*; and *consideration of cultural differences and values*.

For *Changes to communication strategies*, the participants recommended: including fathers in conversations and directing attention to them; taking time in conversation and avoid rushing patients; adjusting communication strategies to the educational level of fathers, culture, values, comprehension level and interest; focusing on positive aspects; being practical and outlining how fathers can help; listening actively to observations of fathers; and clearly outlining expectations from the start.

Several suggestions on how the process of *Assessment*, *treatment*, and *follow-up* can be improved emerged and included: involving fathers in the intervention and assessment sessions as much as possible, providing visual supports in home programs, following-up with fathers if they are absent, and modifying the assessment setting (by including a double-sided mirror) to alleviate the stress related to the presence of HCP during an assessment conducted *via* observation (e.g., playtime, parent-child interaction, feeding). *Considering both parents' agendas* and *Accommodating fathers' schedules* to promote their presence at medical visits was also reported as one solution. The HCPs also suggested the need to be *Sensitive to cultural differences* and values during interactions and adjust their approach accordingly.

Solutions related to *Education and awareness* were reported by the HCPs only (100%) and included developing and launching: (1) formal education curriculum for trainees with a focus on family-centered care and particularities related to fathers; (2) continuing education material for practicing HCPs in the field of childhood disability, outlining current issues and how to bridge them; and (3) material targeting fathers of children with disability, with an aim to increase their awareness about potential positive outcomes of their involvement and engagement in the healthcare and upbringing of their child.

Changes to *Healthcare services* at large (e.g., improving access, transparency, and visibility of information; medical records, and administration) were proposed entirely by the father-participants (100%). In addition, modifications to *how fathers are portrayed* in the media and in the healthcare system (e.g., program titles) were recommended:

> *HCP13: "So I think also that when we see children represented in the media, they are with mums, and then when we see children with disabilities represented in the media they are with mums and then like if we even manage to get a representation of a dad, we don't often see pictures and videos of dads with kids with disabilities. And if we do, we do not see a spectrum of dad abilities – we get hero- dad. We do not see ordinary dads doing the ordinary things that come with kids with disabilities. I think representation matters and representation of dads who maybe are not superheroes, maybe they are just regular dads who play in regular ways. When dads exist in the media with kids, they are either amazing at that play or they are described as amazing even when they are just doing their jobs. I think we hit both sides of that so I think dads can either get intimidated, like if I am not YouTube worthy, I best not jump in. Or yeah just they don't know that people that look like them can be a part of the picture."*
> *HCP12: "[What was helpful is] for example, our toddler group is called 'Moms and Tots' and recently we changed it to 'Parents and*

TABLE 3 | Solutions to improve interactions and parent–professional relationships between fathers of children with disabilities and HCPs.

Theme n = number of utterances % of Solutions	Subtheme n = number of utterances % of Theme	Sub-subtheme	Description	Father's utterances n (% of total)	HCPs' utterances n (% of total)	Total utterances n (% of Theme)
Solutions for HCPs $N = 46$ 65.7%	*Communication strategies* $N = 20$ 43.5%	Inclusion and direct attention	To include fathers in conversations and direct attention to them in interactions.	1 (12.5)	7 (84.5)	8 (40.0)
		Time	To take more time when interacting with fathers.	1 (33.3)	2 (66.7)	3 (15.0)
		Adjustment, individualization	To adjust communication strategies to father's educational level, culture, values, comprehension level, and interests.	1 (33.3)	2 (66.7)	3 (15.0)
		Positive focus	To focus on strengths and positive aspects.	2 (100)	0 (0)	2 (10.0)
		Practicality	To make conversation practical for fathers and outline how can they help, be educational.	1 (50.0)	1 (50.0)	2 (10.0)
		Active listening	To listen actively to fathers and their observations.	0 (0)	1 (100)	1 (5.0)
		Clear expectations	To outline expectations from the start.	0 (0)	1 (100)	1 (5.0)
			Total	**6 (30.0)**	**14 (70.0)**	**20 (100)**
	Assessment, treatment and follow-up $N = 12$ 26.1%	Involvement in intervention session	To involve fathers in interventions sessions.	1 (16.7)	5 (83.3)	6 (50.0)
		Involvement in assessment sessions.	To involve fathers in assessments and screening sessions.	0 (0)	2 (100)	2 (16.6)
		Visual supports for home programs.	To develop visual supports for home programs and include pictures of male figures.	2 (100)	0 (0)	2 (16.6)
		Follow-up	To follow up with fathers if they are absent from the session by providing updates and keeping them informed.	0 (0)	1 (100)	1 (8.3)
		Assessment setting modification	To modify assessment setting by including a double-sided mirror.	0 (0)	1 (100)	1 (8.3)
			Total	**3 (25.0)**	**9 (75.0)**	**12 (100)**
	Appointments and scheduling $N = 9$ 19.5%	Agenda	To consider both parents' agendas and accommodate father's schedule for him to be present.	1 (14.3)	6 (85.7)	7 (77.7)
		Crucial timepoints	To insist on seeing both parents at crucial timepoints (initial assessment, diagnosis, planning for treatment, re-assessments).	0 (0)	2 (100)	2 (22.2)
			Total	**1 (11.1)**	**8 (88.8)**	**9 (100)**
	Fair consideration of fathers vs. mothers $N = 4$ 8.6%	-	HCP to offer fair attention to fathers and mothers by considering them as equal parent-partners.	1 (25.0)	3 (75.0)	4 (100)
			Total	**1 (25.0)**	**3 (75.0)**	**4 (100)**
	Consideration of cultural differences and values $N = 1$ 2.1%	-	HCP to consider cultural difference and values that might be at play (their own vs. patient's) and be sensitive to those factors when interacting with families.	0 (0)	1 (100)	1 (100)

(Continued)

TABLE 3 | Continued

Theme *n* = number of utterances % of Solutions	Subtheme *n* = number of utterances % of Theme	Sub-subtheme	Description	Father's utterances *n* (% of total)	HCPs' utterances *n* (% of total)	Total utterances *n* (% of Theme)
			Total	**0 (0)**	**1 (100)**	**1 (100)**
Education and awareness *N* = 11 15.7%	*Formal education curriculum for trainees* *N* = 6 54.5%	-	Refers to the development and inclusion of an educational module (for trainees) that is specific to family-centered care in the formal curriculum within the health-sciences programs (pediatric-related sections).	0 (0)	6 (100)	6 (54.5)
	Continuing education for HCPs *N* = 4 36.4%	-	Refers to the development and launch of a continuing educational module that is specific to family-centered care for practicing clinicians in the field of childhood disability.	0 (0)	4 (100)	4 (36.3)
	Fathers' awareness *N* = 1 9.0%	-	Refers to the development and launch of knowledge translation material to raise awareness of fathers (and other family members) about their positive contributions to child's health and family's well-being when they are actively involved and engaged in child's healthcare and upbringing.	0 (0)	1 (100)	1 (9.1)
			Total	**0 (0)**	**11 (100)**	**11 (100)**
Healthcare services and media *N* = 11 15.7%	*Access* *N* = 5 45.4%	-	Refers to improving access, transparency and visibility of information and services.	5 (100)	0 (0)	5 (45.4)
	Public relations (media, titles of groups/programs) *N* = 4 36.6%	-	Refers to changing how fathers are portrayed in the media and in materials used by the healthcare settings and modify the titles of groups/program to be inclusive of fathers.	0 (0)	4 (100)	4 (36.3)
	Medical records/administration *N* = 2 18.2%	-	Refers to minimizing administrative and medical record biases (e.g., primary phone number is always that of the mother) to accommodate both parents and promote inclusiveness.	1 (50.0)	1 (50.0)	2 (18.1)
			Total	**6 (54.5)**	**5 (45.5)**	**11 (100)**
Solutions for fathers *N* = 2 2.8%	*Engagement* *N* = 1 50%	-	Refers to fathers to actively engage in conversations during interactions with HCPs.	1 (100)	0 (0)	1 (50.0)
	Awareness *N* = 1 50%	-	Refers to fathers gaining awareness about their potential impacts of involvement in child's healthcare.	0 (0)	1 (100)	1 (50.0)
			Total	**1 (50.0)**	**1 (50.0)**	**2 (100)**
			Grand Total	**18 (25.7)**	**52 (74.3)**	**70 (100)**

> *Tots.' I find it makes a difference, the fathers that started to get included they kept asking: Why it is called 'Moms and Tots,' and so little things like that…"*

Lastly, solutions *Targeting fathers* emerged and included: increasing engagement and awareness of fathers about their potential impacts of engagement in the healthcare of their child.

DISCUSSION

To enhance healthcare-related interactions and experiences for fathers of children with developmental disabilities, the goal was to explore the barriers to and facilitators of positive and empowering interactions and parent–professional relationships, from the perspectives of fathers of children with developmental disabilities and HCPs. The participants of this study reported numerous impacts of positive interactions and involvement of fathers in the healthcare of their children. Also, we determined several influential factors (obstacles vs. enablers) that are at play during parent–professional interactions. We also outlined practical solutions that can be implemented in various clinical settings with the aim of improving interactions and parent–professional relationships, as well as promoting family-centered care.

In this study, the fathers reported to be moderate to very much involved in the healthcare of their child (e.g., arranging healthcare services and being present during medical visits) and other activities (e.g., home program, feeding, exercises). This is congruent with results from previous qualitative studies on fathering children with disabilities (19–21). Nonetheless, the HCP-participants conveyed that fathers are only somewhat involved in those activities. The discrepancy in findings between the two study groups could be attributed to selection and response biases. To explain, the interviewed HCPs have experience with numerous fathers of children with disabilities with different engagement levels (involved vs. not involved). On the other hand, the fathers who participated voluntarily in this study are parents who are moderate to very much engaged in the healthcare and upbringing of their child. However, previous research has suggested that, resulting from cultural beliefs about parenting roles in the family, some fathers of children with disabilities may choose to be less involved with their children. It is common for the mother to undertake the role of primary caregiver and the care from the father related to the child becomes optional (22). Similarly, we also found that cultural factors, such as the traditional beliefs about the role of the father in the upbringing and healthcare of the child, represented 20.7% of all reported barriers and were predominantly conveyed by the HCPs (88.8% of the time). Furthermore, the fathers reported that although they are overall satisfied with the interactions they have had with HCPs in the past, they report that HCPs are only sometimes attentive to them during interactions. This finding resonates with previous research where many fathers report feeling "invisible" during interactions with service providers in relation to the care of their child (23, 24).

Personal factors related to fathers (e.g., their level of engagement/involvement, personality traits, denial vs. acceptance, difficulty opening up about issues) accounted for most of the obstacles or enablers to optimal interactions and parent–professional relationships. Those factors may originate from and be further exacerbated by the high stress levels and mental health issues potentially experienced by fathers of children with disabilities. For example, Giallo et al. (25) demonstrated that in fathers of children with disabilities, there were high rates of symptoms of depression and stress, with nearly 8% reporting severe to extreme symptoms. A recent Canadian study using population-level administrative data from the Ministry of Health compared the mental health of parents of children who have a DD with the mental health of parents of typically developing children. They reported that diagnosed mental health issues were the most prevalent health issues among parents of children with disabilities (including fathers at 60%) (26). The findings of this study regarding external factors, such as supportive vs. hindering work conditions, resonate with previous research suggesting that parents of children with disability experience different stressors, capacity to work, and patterns of pastimes compared with parents of typically developing children (27). It is, therefore, important for HCPs to be aware of those elements and to be sensitive to potential mental health challenges and lived stressors when interacting with fathers of children with disabilities.

Further, the father's denial vs. acceptance of the child's condition, strengths, and challenges was identified as an important influential factor by both study groups. Comparatively, a recent study explored experiences of fathering children with autism spectrum disorder (19). In this study, fathers described their experiences as a "path toward acceptance." Hence, we propose that HCPs support fathers in their journey toward acceptance and this might lead to enhanced connections and experiences.

Similar to previous findings, where fathers report feeling overwhelmed and overshadowed during interactions with HCPs (22, 24), we found that factors, such as family dynamics (e.g., overpowering partner), factors related to HCPs (e.g., focusing on negative aspects, lack of knowledge about cultural factors, lack of attention to fathers), and time limitation, where fathers feel rushed through the medical visits, are hindering interactions and parent–professional relationships. The study participants also reported several issues with the healthcare system in general (e.g., long waiting times, lack of visibility of services and transparency), and that previous negative experiences with the healthcare system shaped them into being more vigilant, defensive, or protective. Analogous results have been reported in the past, where fathers of children with disabilities saw themselves as "advocates fighting obstructive services to access appropriate care" (19). On the other hand, when HCPs provide explanations, take their time during interactions, implement family-centered approaches in care, demonstrate intent to involve fathers, advocate for the family, and engage fathers whenever possible, it enables interactions and builds and maintains parent–professional relationships.

Finally, in line with identified barriers and facilitators, several practical solutions emerged. These solutions can be easily implemented in the clinical settings of today to enhance the experiences of fathers of children with disabilities and promote helpful and empowering interactions between fathers and HCPs. The majority of the suggestions pertain to communication strategies (e.g., directly including fathers and paying attention to fathers in sessions), changes in clinical practices related to evaluation, treatment, administrative system (scheduling), family-centered care (where both parents are considered equally), and consideration of cultural differences. Access to information and services, as well as how fathers are portrayed vs. not portrayed in the media and within the healthcare system should also be evaluated and adjusted as needed. We propose that future research should focus on promoting the development and launch of knowledge translation tools to include educational materials to increase knowledge and awareness of trainees, practicing clinicians, and parents about fathers of children with disabilities.

There are limitations in this study, such as the small sample size of father-participants, potentially limiting the generalizability of the findings. Nonetheless, the data analysis showed saturation of ideas with the included participants. To the advantage of the authors, the sample also included fathers of children from a wide age range and variability in diagnoses, single and married individuals, and of different educational backgrounds. The use of surveys and interviews enabled us to triangulate perspectives through mixed methods, allowing a richer understanding of the experiences of fathers and HCPs. In addition, given the use of patient-oriented research strategy, the study greatly benefited from the inclusion of two father-advisors on the research team, boosting the relevance and suitability of measurement strategies and interpretation of findings.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The Research Institute of the McGill University Health Center and University of British Columbia. The participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

TO drafted the first version of the manuscript. All the authors developed the study protocol, reviewed the manuscript, provided modifications, and suggestions where appropriate.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.709262/full#supplementary-material

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REVIEW
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A Narrative Review of Function-Focused Measures for Children With Neurodevelopmental Disorders

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Clinical measures in health and rehabilitation settings are often used to examine child functioning to better support the diverse needs of children with neurodevelopmental disorders (NDD) and their families. The WHO's International Classification of Functioning, Disability, and Health (ICF) framework reflects a focus of health beyond biomedical deficits, using the concept of functioning to create opportunities for measurement development involving this construct. In the measures developed in the field of childhood NDD, it is unclear whether and how these tools measure and incorporate the ICF framework and its domains within health care contexts. Understanding how these measures utilize the ICF will enable researchers and clinicians to operationalize function-focused concepts in studies and clinical practice more effectively. This narrative review aims to identify and describe function-focused measures that are based on the ICF for children with NDD, as described in the peer-reviewed literature. This review used a systematic search strategy with multiple health-focused databases (Medline, PsycInfo, EMBASE, EMCARE), and identified 14 clinical measures that provide direct support for children (aged 0-21) with NDD in pediatric health (and other) settings. Results described the measures that were primarily developed for three main *diagnostic populations* [cerebral palsy, autism spectrum disorder, and communication disorders]; had varying *contextual* use (clinical-only or multiple settings); and for which authors had conducted *psychometric tests* in the measure's initial development studies, with the most common being content validity, interrater reliability, test-retest reliability. Participation (79%, $n = 11$) & Activities (71%, $n = 10$) were the most common ICF domains captured by the set of measurement tools. Overall (71%, $n = 10$) of the identified measures utilized multiple ICF domains, indicating that the "dynamic nature" of the interactions of the ICF domains was generally evident, and that this result differentiated from "linking rules," commonly used in research and clinical practice. The implications of these findings suggest that clinical measures can be an effective application of the ICF's defined concepts of functioning for children with NDD.

Keywords: children, neurodevelopmental disabilities, clinical measures, ICF, NDD

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INTRODUCTION

Neurodevelopmental disorders (NDD), as defined by the Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5), refer to a group of conditions that present during a child's early developmental period and are characterized by developmental deficits that may create challenges in the child's personal, social, academic, or occupational functioning (1). Common examples of NDD include autism spectrum disorder (ASD), communication and/or language disorders, attention deficit/hyperactivity disorder, motor disorders (including cerebral palsy [CP]), learning disorders, and developmental coordination disorder (1). The prevalence rate for NDD in developed countries range from 7 to 14% of all children (2). Children with NDD may experience challenges in different environments, potentially impacting their functioning within academic settings (school), daily living skills (home), and the broader community (3–7). The DSM-5 describes these challenges as a symptom of excess, deficit, or delay in key aspects of child functioning, especially when considering the achievement of expected developmental milestones (1).

Historically, biomedical models and thinking have greatly influenced clinical practice, including the field of childhood disability (8, 9). This traditional way of thinking focused on the attributes of a child's deficits and limitations, for diagnostic purposes and to treat aspects of the child's "disability" (10, 11). In 2001, the International Classification of Functioning, Disability, and Health (ICF) – a contemporary conceptual framework – challenged these practices and highlighted the paradigm shift to think beyond the biomedical model to an integrated biopsychosocial model of human functioning and disability (8). This biopsychosocial model emphasizes that individuals with disabilities have needs that extend beyond the medical scope of practice, and are often broad-based in nature within social, educational, and functional settings (12).

As shown in **Figure 1**, the four key domains of the ICF are: *body function* & *structures* (functioning at the level of the body); *activities* (functioning at the level of the individual); *participation* (functioning of a person as a member of society); and *contextual factors* (personal and environmental factors that can exist as facilitators or barriers) (8). The ICF framework defines *functioning* as an umbrella term to describe the interactions of these four domains, examining the positive or neutral aspects occurring between the individual's health condition(s) and their context (8). "Disability" is an alternate umbrella term used to describe the interactions of an individual's impairments, activity limitations and participation restrictions, examining the negative aspects of the *interaction* between the individual's health condition(s) and their context (8).

This ICF framework depicts the interactive and non-linear nature of the core domains, establishing that these conceptual domains are not independent when examining functioning and/or disability. The framework is representative of the biopsychosocial perspective, as it recognizes how the influences of physical, psychological, and social factors within "functioning and disability" can be understood from the viewpoint of the individual with respect to their health condition (13, 14). Without focusing on single descriptors to label a child's functional abilities, this framework utilizes a holistic approach that still highlight the nuances in the interactions of the different elements that build a child's functional profile (14, 15). This framework indicates a paradigm shift in the ways that researchers and clinicians understand disability, as it provides a multidimensional perspective that both classifies functioning independently from the individual's diagnosis and views disability as product of person-environment interactions (12, 15, 16).

Children with NDD exhibit a wide range of levels of functioning within and between their diagnostic groups (17). In addition to the ICF framework, the field of NDD has seen growth in the concept of functioning that takes into consideration the heterogeneous level of abilities within diagnostic populations that extends across NDD (10). For example, within ASD literature, the concept of *neurodiversity* views neurological differences as inherent human variation, rather than as a disorder, and celebrates the individuality of a person – regardless of their capabilities (18, 19). This change in thinking in the field of ASD has had a great influence in promoting various abilities and child differences within ASD interventions, including the language that is being used to describe the diagnosis (19, 20).

Similar concepts of functioning started in the field of CP, in relation to interventions in pediatric rehabilitation. Rather than using the traditional approaches of CP that attempt to normalize movement patterns and minimize the development of secondary impairments, there is an increased emphasis on enabling the child to master various tasks and participate in different activities (21, 22). Over the last 20 years, in the field of CP, clinical care and research have examined child functioning as it relates to interacting contextual factors (22, 23). Although the needs and abilities of children with NDD (and their families) are heterogeneous, everyday functioning is continually regarded as an important outcome to families (24).

Examining clinical contexts in particular, there is a notable emphasis of functioning in the ICF, as this term is often used to describe abilities-focused processes – otherwise commonly referred to as function-focused care (12, 24, 25). Within this type of pediatric care planning, there are typically certain measures used with families to promote child functioning or child abilities.

Although these measures may have the appropriate psychometric testing completed to illustrate their effectiveness in clinical utility, it is also important that there is some consistency with the language that is being used with these measures (20, 26, 27). For example, the terms "function," "functional," and "everyday functioning," are used synonymously in the literature, whereas the ICF's conceptualization of functioning emphasizes it as the complex interactions between the four domains (10). There are various measures that aim to assess concepts related to function, such as adaptive behavior [e.g., Vineland Adaptive Behavior Scales (28), Adaptive Behavior Assessment System (29), Behavior Assessment System for Children (30)]. However, these measures are not based on the ICF, and therefore describe everyday function differently compared to ICF-based measures.

Operationalizing the ICF framework (i.e., its domains and the interactions between them) within measurement tools can create opportunities for the ICF to be widely utilized in clinical

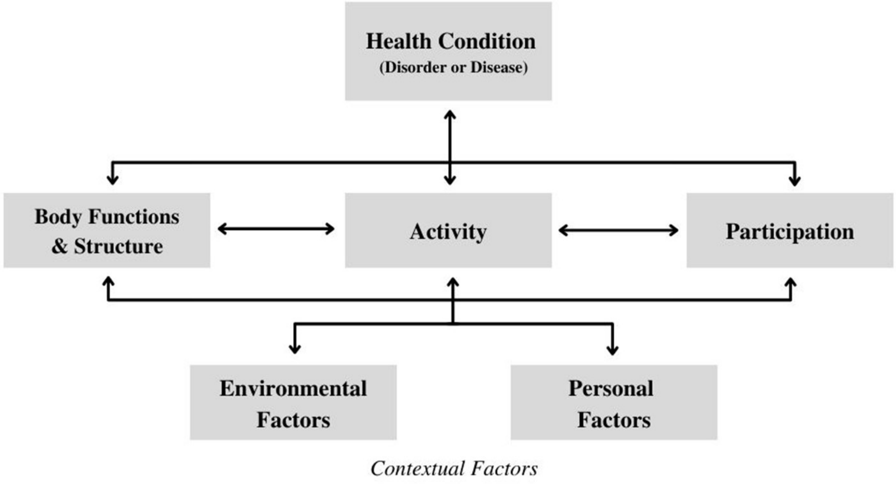

FIGURE 1 | International classification of functioning, disability, and health framework (8).

environments for children with NDD (14). It is unclear how many measures in the field of NDD are developed using the definitions and concepts of the ICF framework. It has been demonstrated in the literature that clinical measures can be mapped on or "linked" to the ICF framework by following a set of established and standardized rules, as described by Fayed et al. (31). With NDD interventions shifting to focus more specifically on strengths and support needs, there is a need for further description of how measures that purport to be function-focused are utilized in clinical systems. The focus of this study is directed toward examining how researchers who have developed ICF-based measures conceptualized their measure, specifically with whether and how the ICF domains were utilized in pediatric clinical contexts and research.

METHODS

We undertook a narrative review and synthesis of the peer-reviewed literature to understand existing function-focused, ICF-based measures that are used with children diagnosed with NDD. A narrative review summarizes and describes previously published information with an interpretation of the contents of different studies using a comprehensive, critical and objective analysis (32, 33). This study was guided by SANRA, the Scale for the Assessment of Narrative Review Articles, specifically by reviewing the six items deemed necessary for a quality review: (1) justification of the review's importance for the reader; (2) review focus/aim(s); (3) description of literature search; (4) referencing; (5) scientific reasoning; and (6) relevant and appropriate endpoint/presentation data (33).

We used a two-stage approach to review the literature. The first stage was to identify original research texts that (a) focused on children (18 and younger) diagnosed with NDD defined by DSM-5, and (b) referenced the ICF framework. Both criteria needed to be stated within the abstract of the study. Initial keywords were generated for each conceptual category of the research aim with the guidance of a trained librarian in a health sciences library to form the search strategy. Keywords were identified within three categories (NDD diagnosis, child [age range 0–18], and ICF), and were used to search the following databases: Medline, PsycInfo, EMBASE, and EMCARE. Search terms were developed and customized for each database. Abstracts were then screened to identify whether any measures were used in an intervention study and/or discussed in the literature; we also required the clinical measure to be the focus of the abstract. Searches were restricted to both English language journals and publication date (2002-April 2021), as selected papers were required to be published post-publication of the ICF in 2001. Studies were excluded if the aim was to translate the measure to explore psychometric properties within an alternate language/country/context. Measures that were used within indirect care (i.e., measures that focused on data collection and/or inter-professional collaboration) were also excluded. Lastly, secondary studies (i.e., systematic reviews, scoping reviews) as well as editorials and commentaries were excluded. The titles and abstracts of the resulting articles from the database search were exported to Covidence (34), a reference managing software. Duplicate records were then deleted using the software.

The second stage required full-text screening to identify whether select measures were ICF-based, and to identify whether the study reported the development of the measure. If a study described an ICF-based measure but was not the original paper of the measure's development, hand-searching was conducted to retrieve the original article describing its development. Hand-searching for original articles was accomplished by looking at the reference lists of the indexed articles that had described the use of these measures within their abstracts. This task was also completed using Covidence (34).

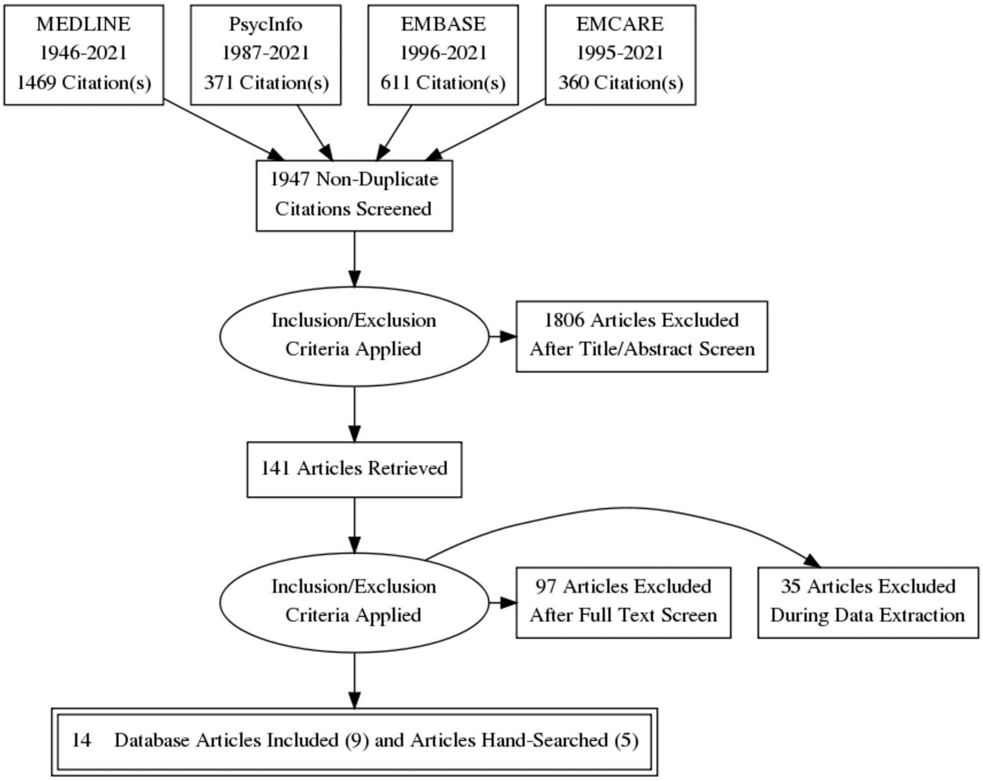

FIGURE 2 | PRISMA (40) flowchart of search strategy.

This study used a matrix to extract key details including age ranges, context(s), diagnosis sample, as well as descriptions and psychometric properties of the measures described. Details and descriptions of these measures were determined by using its original development article. After the characteristics of the included measures were extracted, the original studies of the measures were analyzed again to extract ICF-related details, specifically regarding the ICF domain(s) that were prioritized by the measure and how this framework influenced the measure's initial conception. Measures were categorized by using the definitions of the four domains of the ICF framework (body structures and function, activities, participation, and contextual factors).

RESULTS

The initial search identified 2811 published abstracts. After duplicates were removed, 1947 papers remained. These papers were reviewed by title and abstract with the first set of inclusion criteria, resulting in 141 potentially relevant studies. For the second stage, full-text versions of these studies were obtained and reviewed to assess whether they fit the second set of inclusion criteria, at which time 97 studies were excluded. Studies were excluded mainly for having a non-relevant focus—not focused on an ICF-based measure providing direct support for children with NDD; focusing on a non-pediatric sample; or being based on secondary data. The 44 papers that remained included 9 studies that described the initial development of an ICF-based measure, and 35 articles that described the use of ICF-based measures but were not the measure's original development paper, for which hand-searching was then necessary.

From the 35 articles, five additional ICF-based measures for children with NDD were identified and included; these comprised of four additional studies (35–38), and one manual (39). In total, 14 initial development studies describing 14 individual measures were included. This information is summarized using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart in **Figure 2** (40).

The 14 measures originated in seven countries including Canada, US, UK, Australia, Sweden, Switzerland, and Taiwan. These measures were predominately described as either assessment and/or outcome measures (64%, $n = 9$) or classification systems (36%, $n = 5$), and could be utilized in various contexts including home, community, educational, and clinical environments. The most common diagnosis was CP (50%, $n = 7$), followed by non-diagnostic/multiple diagnoses (29%, $n = 4$), ASD (14%, $n = 2$), and communication disorders (7%, $n = 1$). Age applicability of these measures ranged from 0 to 21 years. The diagnosis sample, age groups, and brief descriptions are reported in **Table 1**. The common characteristics of these measures are also described.

TABLE 1 | General description and characteristics of ICF-based clinical measures.

Measure acronym	Measure full name and citation	Type of measure	Country of origin	Primary context(s)	Diagnosis sample	Age range	Construct of interest	Brief description
ICF-CS	ICF Core Set (41)	Standardization for Assessment and Description	Switzerland	Multiple (i.e., clinical, home, educational, community)	Multiple versions with different diagnoses (ASD, ADHD, CP)	Multiple versions with different age ranges	Functional Abilities	This measure uses select categories from the ICF classification to describe relevant information in regards to an individual's level of functioning; this helps facilitate a systematic and comprehensive system for either a specific health condition or health context (41). There are two versions of this measure: ICF Comprehensive Core Sets and ICF Brief Core Sets (41).
GMFCS – E&R	Gross Motor Function Classification System Expanded and Revised (35)	Classification System	Canada	Clinical	Cerebral Palsy	0–18 years	Gross Motor Function	A 5-level classification system that describes gross motor function for children and youth with CP, specifically focused on self-initiated movement when a child sits, walks, and/or uses a wheeled mobility device (35).
MACS	Manual Ability Classification System (42)	Classification System	Sweden	Clinical	Cerebral Palsy	4–18 years	Manual Ability	Developed from the GMFCS, this 5-level classification system examines typical manual performance of children with CP, specifically in regards to a child's ability to handle objects (i.e., assistance needs, potential adaptations required, quantity/quality of performance) (42).
BFMF	Bimanual Fine Motor Function (43)	Classification System	Sweden	Clinical	Cerebral Palsy	Not specified	Fine Motor Function	A 5-level classification system that examines fine motor function in children with CP, specifically in regards to a child's ability to grasp, manipulate, and hold objects for each hand (43).
CFCS	Communication Function Classification System (44)	Classification System	USA	Multiple (i.e., clinical, home, educational, community)	Cerebral Palsy	2–18 years	Communication	A 5-level classification system used by clinicians for children with CP, to classify and understand the patterns of a child's performance in everyday communication effectiveness with a partner (44).
ACSF:SC	Autism Classification System of Functioning: Social Communication (36)	Classification System	Canada	Multiple (i.e., clinical, home, educational, community)	ASD	3–5 years	Social Communication	A 5-level classification system that provides a simplified method to describe social communication functioning for preschool children with ASD (36). This measure provides parents and service providers with an understanding of the potential differences in social communication abilities based on a child's capacity and typical performance within different contexts (36).
GOAL	Gait Outcomes Assessment List (45)	Assessment Measure	Canada	Clinical	Cerebral Palsy	Not specified	Gait Priorities	An assessment measure that evaluates gait priorities and functional mobility for ambulant children with CP, addressing the spectrum of needs and/or goals of these children and their caregivers (45).
FOCUS®	Focus on the Outcomes of Communication Under Six (46)	Outcome Measure	Canada	Clinical	Communication Disorders	0–6 years	Communication	An outcome measure that evaluates change in communicative-participation, examining 'real world' changes in preschool children's communication abilities (46).
QYPP	Questionnaire of Young People's Participation (47)	Assessment Measure	United Kingdom	Clinical	Cerebral Palsy	13–21 years	Participation	A 45-item questionnaire assessing participation frequency across multiple domains for children and adolescents with cerebral palsy (47).

(Continued)

TABLE 1 | Continued

Measure acronym	Measure full name and citation	Type of measure	Country of origin	Primary context(s)	Diagnosis sample	Age range	Construct of interest	Brief description
MEVU	Measure of Early Vision Use (48)	Assessment Measure	Australia	Clinical	Cerebral Palsy	Not specified	Vision	A measure that examines typical performance with 'how vision is used' during a child's everyday activities, interactions and environments (48).
CAP-HAND	Children's Assessment of Participation with Hands (37)	Assessment Measure	Australia	Multiple (i.e., clinical, home, educational, community)	No specific diagnosis	2–12 years	Participation	A parent report questionnaire, examining upper limb abilities across disorders, as well as the extent to which children participate in life situations (with a focus on hand use) (37).
CAPE & PAC	Children's Assessment of Participation and Enjoyment and Preferences for Activities of Children (39)	Assessment & Outcome Measure	Canada	Multiple (i.e., clinical, home, educational, community)	No specific diagnosis	6–21 years	Participation and Activity Preferences	Together, CAPE & PAC are self-report measures that examine children's participation and activity preferences within six dimensions of activity (39). CAPE documents the extent to which children with or without disabilities participate in everyday activities outside of their mandated school activities. PAC examines children's specific activity preferences (39).
-	ICF-CY Based Questionnaire (49)	Assessment Measure	Taiwan	Clinical	ASD	3–6 years	Functional Profile	This measures comprises of 118 items using the ICF-CY structure to evaluate the functional profiles of preschool children with ASD (49).
PEM-CY	Participation and Environment Measure for Children and Youth (38)	Outcome Measure	USA	Multiple (i.e., clinical, home, educational, community)	No specific diagnosis	5–17 years	Participation and Environment	This parent-reporting survey allows parents, researchers, and service providers to better understand a child's participation patterns in home, school, and community settings, by studying both participation and environmental factors at the same time (38).

TABLE 2 | Psychometric properties described in the measures' initial development.

Measure acronym	Development study	Diagnosis sample	Description of psychometric properties in development article
ICF-CS	A guide on how to develop an International Classification of Functioning, Disability and Health Core Set (41)	Multiple versions with different diagnoses (ASD, ADHD, CP)	Not included in the development article.
GMFCS – E&R	Development of the Gross Motor Function Classification System for cerebral palsy (35)	Cerebral Palsy	Not included in the development article.
MACS	The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability (42)	Cerebral Palsy	External construct validation process was initiated, involving rehab professionals within pediatric rehabilitation and parents of children with CP (42). Interrater reliability was conducted using testing between parents and therapists (42).
BFMF	Neuroimpairments, activity limitations, and participation restrictions in children with cerebral palsy (43)	Cerebral Palsy	Not included in the development article.
CFCS	Developing and validating the Communication Function Classification System for individuals with cerebral palsy (44)	Cerebral Palsy	The second and third phases of the measure's development focused on revision and validation using nominal group studies and Delphi surveys (content validity) (44). The fourth phase measured interrater reliability among clinicians and parents as well as test-retest reliability (44).
ACSF:SC	Developing a classification system of social communication functioning of preschool children with autism spectrum disorder (36)	ASD	Interrater reliability reported good for parents and very good for professionals (36). Content validity of level descriptions and ratings were trialed by participants in each stage of measure development using surveys (36).
GOAL	The Gait Outcomes Assessment List: validation of a new assessment of gait function for children with cerebral palsy (45)	Cerebral Palsy	Concurrent validity was assessed comparing the GOAL with two related valid and reliable assessments of motor function (45). Further studies will be required with larger cohorts to assess validity and reliability of the GOAL in different populations (45).
FOCUS®	Development of the FOCUS (Focus on the Outcomes of Communication Under Six), a communication outcome measure for preschool children (46)	Communication Disorders	Parents completed the Pediatric Quality of Life Inventory (PedsQL) at the start and completion of treatment to evaluate FOCUS' content validity (46). Parents and clinicians completed the FOCUS measure twice within a 1 week period for test-retest reliability (46).
QYPP	The Questionnaire of Young People's Participation (QYPP): a new measure of participation frequency for disabled young people (47)	Cerebral Palsy	Test-retest reliability was examined by intra-class correlations using a two-way mixed model; results were comparable with other participation measures (i.e., GMFCS, MACS) (47). Using a rigourous expert review of the measure's item pool, content validity was maximized; known-groups (discriminant) validity was also supported (47).
MEVU	Measure of Early Vision Use: development of a new assessment tool for children with cerebral palsy (48)	Cerebral Palsy	Not included in the development article.
CAP-HAND	Development and Psychometric Evaluation of a New Measure for Children's Participation in Hand-Use Life Situations (37)	No specific diagnosis	Evidence for construct validity was established using Rasch analysis. Differences in summary scores of each domain between children with and without disabilities were also significant (37). Test-retest reliability using ICCs of the measure was moderate-high, except for a single dimension scale. Internal consistency varied across the dimensions, providing preliminary evidence for construct validity and reliability (37).
CAPE & PAC	Children's Assessment of Participation and Enjoyment and Preferences for Activities of Children Manual (39)	No specific diagnosis	Information from the measure's longitudinal study was used to examine the technical characteristics of the CAPE and PAC (39). The data provided evidence of reliability and validity of the CAPE and PAC (39).
ICF-CY Based Questionnaire	ICF-CY based assessment tool for children with autism (49)	ASD	This measure has evidence of good interrater reliability, expert (construct) validity, and reflects the functional profile of preschool children with autism (49). Further testing is required to confirm other psychometric characteristics (49).
PEM-CY	Development of the participation and environment measure for children and youth: conceptual basis (38)	No specific diagnosis	Not included in the development article.

Assessment (and Outcome) Measures

The following nine ICF-based measures have a primary focus on the assessment of a specific construct of interest: ICF Core Sets (ICF-CS) (41), Gait Outcomes Assessment List (GOAL) (45), Focus on the Outcomes of Communication Under Six (FOCUS®) (46), Questionnaire of Young People's Participation (QYPP) (47), Measure of Early Vision Use (MEVU) (48), Children's Assessment of Participation with Hands (CAP-HAND) (37), Children's Assessment of Participation and Enjoyment & Preferences for Activities of Children (CAPE & PAC) (39), the ICF-CY Based Questionnaire (49), and the Participation and Environment Measure for Children and Youth (PEM-CY) (38). These types of measures describe details of functioning, can observe and evaluate a child's abilities and limitations within the construct of interest (otherwise referred to as outcome measures—a subset of assessment measures), and in some cases, it may be used to predict within-person change over time (26). These assessments can be completed by various individuals that are familiar with and/or are knowledgeable about the child's competencies within their daily routines, including caregivers, clinicians, and teachers (39). With the conceptual grounding of the ICF, these measures can provide a comprehensive and clinically useful understanding of a specific phenomenon, which can then be used for various applications within research and practice (39).

Classification Systems

The remaining five ICF-based measures are classification systems that can be used for children with NDD: Gross Motor Function Classification System Expanded & Revised (GMFCS-ER) (35), Manual Ability Classification System (MACS) (42), Bimanual Fine Motor Function (BFMF) (43), Communication Function Classification System (CFCS) (44), and Autism Classification System of Functioning: Social Communication (ACSF:SC) (36). The GMFCS-ER (35), MACS (42), BFMF (43), and CFCS (44) each individually describe functioning in children with CP based on specific constructs (i.e., gross motor function, manual ability, fine motor function, and communication), and the ACSF:SC (36) describes social communication functioning in children diagnosed with ASD. In these classification systems, level I typically describes child functioning with the highest level of ability in that aspect of functioning, whereas levels IV-V typically describe child functioning with more significant limitations (43). The five levels in these systems are ordinal, describing different levels of a child's abilities for a specific construct (36). It is important to note that the differences between these levels are not equal, as these systems provide a simplified guide for families and clinicians to communicate level of functioning within the clinical process (43).

Psychometric Properties of Development Studies

The studies in which these measures were first established were published between 2002 and 2021. Almost all measures were initially published in journal articles (93%, $n = 13$), with one measure [CAPE & PAC (39)] described in a manual format. Most studies of these measures (64%, $n = 9$) provided some psychometric testing information during the measure's development. The most common forms of testing include content validity, interrater reliability, and test-retest reliability (29%, $n = 4$). Other types of psychometric testing include various types of construct validity testing, such as discriminant validity, expert validity, and concurrent validity (each 7%, $n = 1$) or were generally described as construct validity (14%, $n = 2$). The CAPE & PAC (39). Manual did not specify the type of reliability and validity results (see **Table 2**).

ICF Domains of Measures

To understand the role of the ICF framework in the conception of these clinical measures, it was important to analyze what ICF domain(s) were prioritized, and the specific foundational concepts from the ICF framework during the initial development process (see **Table 3**). All listed measures included at least one domain of interest, and the ICF-CS (41), GOAL (45), and ICF-CY Based Questionnaire (49) using all four ICF domains. The most common domain across measures was Participation (79%, $n = 11$), followed by Activities (71%, $n = 10$), Contextual Factors (43%, $n = 6$), and Body Structures and Function (29%, $n = 4$). Seventy one percent of the measures ($n = 10$) used more than one domain of the ICF.

DISCUSSION

This study is the first of which we are aware to identify ICF-based clinical measures for children with NDD. We have reported the psychometric properties and characteristics of 14 measures that are grounded in the ICF framework, using the information gathered from the initial development studies. We also identified the prominent ICF conceptual domain(s) that these measures represent, and the extent to which the framework was captured, including its interactive nature. There may be more ICF-based measures for this population that exist outside the clinical context (i.e., educational-based measures) and some of these tools may be applicable to other settings; however, the intent of this study was to examine how ICF-based clinical measures were operationalized in practice. Therefore, only health-focused databases were consulted.

The initial development studies for the selected measures included varying levels and types of psychometric properties conducted and described. Some studies [ICF-CS (41), GMFCS-ER (35), BFMF (43), MEVU (48), and PEM-CY (38)] placed emphasis on the process that the research team experienced when developing the measure, rather than describing specific psychometric characteristics of their measure. These studies had concurrent publications that described the conceptual processes and psychometric testing separately. The remaining studies combined psychometric testing with the measure's development process. The most common psychometric tests that were completed were interrater and test-retest reliability as well as content validity. It is important for clinical instruments to demonstrate good psychometric properties (27), and 64% of the measures were introduced with some form of psychometric testing conducted. These results provide a descriptive overview of the function-focused tools developed in the field of childhood

TABLE 3 | ICF domains prioritized in the development of the measure.

		ICF domains			**Total domains**	**How is the ICF described overall?**
	Body structures and function	**Activities**	**Participation**	**Contextual factors**		
ICF-CS	✓	✓	✓	✓	4	This instrument selects essential categories that cover each component of the ICF.
GMFCS – E&R		✓		✓	2	"Our group's perspectives have evolved and been shaped considerably by the World Health Organization's (WHO) International Classification of Functioning, Disability and Health (ICF) […] The basic ideas concerning capacity and performance were included in the original GMFCS concepts but have been sharpened considerably with the publication of the ICF" [35, p. 251].
MACS		✓			1	"The focus is on manual ability, as defined in the International Classification of Functioning, Disability and Health [...] the classification looks at activities and gives a single 'level' for the collaborative use of both hands when handling objects in daily life" [42, pp549-52].
BFMF	✓	✓	✓		3	"Motor function and learning disability were important predictors for participation restrictions in children with CP. The ICF has the capacity to be a model to help plan interventions for specific functional goals and to ascertain the child's participation in society" [43, p. 309].
CFCS		✓	✓		2	"The purpose of this study was to create and validate the Communication Function Classification System (CFCS) for children with CP, for use by a wide variety of individuals interested in CP. This required a shift from the traditional focus on body structure and function (i.e., assessing components of speech, language, and hearing problems), to a focus on activity/ participation, specifically the way in which to classify a person's communication capacity within real-life situations" [44, p. 705].
ACSF:SC		✓	✓		2	"Using the ICF activities and participation framework, resulting autism classifications will focus on how children's differing social communication affects their activities and participation in daily lives" [36, p. 943].
GOAL	✓	✓	✓	✓	4	"Used with gait analysis, the GOAL provides comprehensive assessment across all International Classification of Functioning, Disability and Health domains" [45, p. 619].
FOCUS®			✓	✓	2	"The constructs used in the FOCUS are derived from the ICF framework to measure changes in communication and their impact on participation. The response set in part II of the FOCUS (i.e., "cannot do at all" to "can always do without help") was designed to evaluate the shift from capacity to performance by evaluating the level of assistance required to complete items successfully" [46, p. 51].
QYPP		✓	✓		2	"In developing the new instrument, we differentiated activities from participation at the level of ICF sub-domains, regarding activities as simpler elements of functioning at body level while participation usually includes those sub-domains made up of a number of activity functions and where the result is of intrinsic social and personal importance" [47, p. 501].
MEVU		✓			1	"This new measure is conceptually grounded within the Activity level domain of the International Classification of Functioning, Disability and Health as a measure of a single visual ability construct" [48, p. 1].

(Continued)

TABLE 3 | Continued

	ICF domains				Total domains	How is the ICF described overall?
	Body structures and function	**Activities**	**Participation**	**Contextual factors**		
CAP-HAND			✓		1	"The conceptual frameworks underlying the development of the Children's Assessment of Participation with Hands are the ICF and the ICF-CY, in combination with additional participation definitions/attributes proposed by Coster and Khetani" [37, p. 1046]. ICF provided only an initial framework for the measure's development.
CAPE and PAC			✓		1	"The CAPE and PAC both focus on a subset of the ICF domains of participation and are based on two taxonomies, or classifications, of leisure and recreational participation" [39, p. 7].
ICF-CY Based Questionnaire	✓	✓	✓	✓	4	"The ICF-CY based questionnaire for children with autism comprised 4 domains: body functions, activities, participation and environment" [49, p. 679].
PEM-CY			✓	✓	2	"As defined by the International Classification of Functioning, Disability, and Health (ICF), participation and environment are multidimensional constructs that have been challenging to measure" [38, p. 238]. The ICF provided an initial framework for the measure's development.

NDD, but since potential subsequent psychometric studies were not included in this study, it is difficult to provide comment on the overall rigor of the state of function-focused tools in this field. Future research should examine levels of rigor found in the psychometric properties of the listed tools.

The 14 measures varied in their constructs of interest, age ranges, and diagnoses. These constructs ranged from very specific functional skills [i.e., BFMF: bimanual fine motor function (43)] to broader areas of interest [i.e., ICF-CY-Based Questionnaire: building a functional profile for children diagnosed with ASD (49)]. For age applicability, two measures [CAP-HAND (37) and CAPE & PAC (39)] had expanded upper-age ranges to 21 years old. Many measures focused on specific diagnoses: CP, ASD, and communication disorders. With the broad spectrum of diagnoses involved in DSM-5's definition of NDD, this highlights the need for great representation in other NDD populations. To fill these gaps, measures like the ICF-CS have been continually adapted with subsequent publications to explore the clinical utility of this measure in multiple communities within disability research and practice, including within NDD (50). These diagnosis-based populations include ASD, CP, and ADHD, but the outreach in these diagnosis populations continue to grow today (50, 51).

Furthermore, these results indicated that measures such as the CAP-HAND (37), CAPE & PAC (39), and PEM-CY (38) could be potentially used with any child or youth, regardless of whether they are diagnosed with any condition of NDD, as these measures are not diagnosis-specific. In addition, although a key population of the users of FOCUS® (46) is young children with communication disorders, this tool is designed to address communication needs across all young children with or without disabilities. These findings are important, as they illustrate function-focused measures that examine abilities across diagnoses/conditions – an emerging trend (15). With the various diagnoses categorized within NDD, these measures have a wider scope in reaching different communities, thus creating more opportunities to utilize the concepts of the ICF in clinical and research settings. It is important to note that the ICF is still considered a contemporary framework, and that measures are continuing to be developed, such as the MEVU (48) that was published 1 month prior to conducting the database search for this narrative review.

With the heterogeneity of functional abilities within NDD, and the emerging measures that are being developed without a focus on any specific diagnosis, non-diagnostic ICF-based measures create opportunities for further examination of the continuum of abilities across diagnoses. By doing so, the goal of these measures shifts toward capturing profiles of individual abilities as well as unique differences among children (12). Furthermore, there is some evidence in today's literature that indicates that neurological similarities (i.e., brain structure/activity) that may affect an individual's social communication abilities may exist across diagnoses such as autism and attention deficit hyperactivity disorder; this shows that a child's overall abilities may also overlap across diagnoses (52). This example can be used to challenge the ways in which we can define, diagnose, and "treat" NDD, specifically with how we approach functional perspectives for these populations (53). Measurement tools may still involve neurophysiological processes in their design, but by focusing on a more individualistic foundation, this shift in thinking may better suit the cultural direction of how function-focused care is understood with today's ideas (12). By utilizing these measures across the populations of NDD, we better understand the diversity in the needs of children within their communities. These needs may exist beyond the core domains (i.e., addressing participation needs), and can potentially extend to how we can utilize these measures to improve the overall quality of life of children with NDD (13, 27, 28). As the ICF promotes this understanding that these four domains can build a unique functional profile of a child, the ICF can be seen as a supporting resource within the use of family-centered care to support a child's individualized needs. When this type of care continues to play prominent roles in the design and development of intervention plans for these individuals with disabilities, this may need to be more apparent in the tools that we develop as well. This approach utilizes the biopsychosocial model and will be a more relevant application of the ICF. Furthermore, with children receiving school supports that are often integrated with health services, exploring function-focused measures that are applicable in educational settings, such as the Functional Abilities Classification Tool (54), is also important to examine in future work.

The development studies demonstrated variability in how ICF concepts were foundational within their measures. Some studies explicitly stated that their measure was conceptually based in the ICF whereas others used the ICF to develop their measure's items or constructs. Both the *Participation and Activities* domains were predominately represented throughout all measures whereas *Contextual Factors* and *Body Structures and Function* were not as prominent. The ICF-CS (41) and ICF-CY-Based Questionnaire (49) utilized a holistic approach of the framework rather than focusing on specific domains, and this is evident simply looking at the naming of these tools. Other measures utilize the ICF combined with other frameworks, such as the CAP-HAND (37), that uses the definitions provided by both the framework and what is described by the authors of the PEM-CY (38) to configure a definition for participation that is suitable for the needs of CAP-HAND (37). These results align with the literature, specifically regarding the shift in thinking the ICF proposes: this framework has motivated health service providers to focus beyond "body structures and functions" to include the other roles (i.e., activities, participation) that can impact a child's level of functioning and health (55, 56).

There are different ways that tools and measures interact with the components of the ICF, and some measures can still utilize this framework without using it for its conceptual basis. It is here that the use of the ICF linking rules may become more relevant, as the rules provides an effective method to link meaningful "concepts" of non-ICF-based measures to the most precise category(s) in the ICF framework (57). These concepts could describe health condition, functional activities or any of the contextual factors (31, 57). This "linking process" differentiates from what is being studied in this narrative review, in that we are examining the extent to which ICF-based measures involve the domains of the framework, and the dynamic interactions they capture. This review focused on identifying measures that used the ICF for the initial conception, rather than the measures that have only considered the ICF post-publication or in an "after-the-fact" exploration. With the linking process, the developers of non-ICF based measures undergo the steps required in understanding the ICF to link certain items of their measure to the most relevant domains of the framework (58). However, the use of linking individual concepts of a measure to the ICF framework may not be as effective in demonstrating the interaction between the concepts, especially between activities/participation and contextual factors (59). Of the identified measures, 71% utilized more than one domain of the ICF, often highlighting the various ways in which the nuanced interactions influenced the development of the tool. For example, the PEM-CY (38) evaluates both participation and environmental factors in different settings, and can provide problem-solving strategies to adjust contextual factors within these settings to support further participation (38). As described earlier, the dynamic nature of the interaction of these non-linear domains is one of the most easily identifiable components of the framework. Although there is variability with how these interactions are explicitly described, when a measure is ICF-based and correctly utilizes the framework as a core component, the interactions of the domains are more likely to be inherently captured within the use of the measure.

LIMITATIONS

There are a couple of limitations to report about this study. To begin, there were varying levels of psychometric data that emerged across the initial development studies of the selected measures. While psychometric testing of measures is an ongoing process, we recognize that the original development manuscripts would only have captured psychometric testing at its initial development, and that subsequent studies could have tested additional properties, potentially with other populations of children. We also recognize that the contributors of the development studies may have differed from the original developers of the measure. The aim of this review was to provide an overview of the current ICF-based measures developed for children with NDD, in which we focused on using the development studies as the main sources for this work.

A second limitation relates to study screening process. We selected measures that explicitly used the ICF in the screening of study abstracts in the identified health databases, either in the development study, or in a subsequent published study of the measure that was used to locate the original manuscript. Although there is the potential for other measures to incorporate the ICF framework in some capacity in the development and/or design of their measure, the focus of this work was to identify the measures that explicitly used the ICF and its domains as a foundational element in its work.

CONCLUSION

This narrative review can serve as a potential resource for clinicians/researchers looking to use measures grounded in the ICF framework for children with NDD. These 14 measures can play important roles in creating effective applications of the ICF for exploring child functioning in both research and practice (15, 54, 60). As measures are continuing to be developed using the ICF framework at their foundation, this emerging knowledge can help inform function-focused care. By understanding how function-focused care is operationalized within the measures that we create, we are also able to better understand functioning in clinical care for children with NDD, and whether there are gaps in what is measured. These gaps are also evident in NDD populations where these ICF-based tools are not applied. Future research can explore the expansion of existing ICF-based measures across NDD populations and ages (i.e., adults), in addition to examining measures that impact functioning in other childhood contexts (e.g., home and educational settings).

AUTHOR CONTRIBUTIONS

KS completed the search and wrote the manuscript with support from BD, PR, and MZ. BD supervised the project, with help from PR and MZ as part of the supervising committee. PR, MZ, and BD conceived the original idea. All authors reviewed the results and contributed ideas to the final manuscript.

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Assessing the Quality of Life of Parents of Children With Disabilities Using WHOQoL BREF During COVID-19 Pandemic

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Background: Caring for a child with disabilities is a challenging journey, as the parents must meet greater demands when compared with the parents of children without disabilities. Looking after a child with disablities requires additional financial, social, emotional, and physical resources. Coronavirus 2019 (COVID-19) pandemic has made this even more challenging and impacted the quality of life of parents of children with disabilities.

Methods: The study was an analytical cross-sectional design with two comparison groups: parents of children with developmental disabilities and parents of children without disabilities. The Urdu version of the WHO Quality of Life Measure Abbreviated version (WHOQOL-BREF) was used to measure the quality of life (QoL) among parents. Sociodemographic data were also obtained from the parents.

Results: Parents of children with disabilities had lower overall scores when different domains of QoL were considered (physical health, psychological health, social relationships, and environment) using WHOQoL-BREF. Statistically significant differences were observed in the physical and environmental domains of parental QoL.

Keywords: quality of life, parents, children with developmental disabilities, COVID-19, WHOQoL

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INTRODUCTION

Caring for a child with developmental disabilities is a challenging journey, as the parents must meet greater demands when compared with the parents of children without disabilities. Such parents require additional financial, social, emotional, and physical resources, which may conflict with the competing needs of the other family members. This life-long journey of parents navigating through the medical, developmental, and educational interventions in addition to caregiving responsibilities affects their quality of life (QoL) (1).

Quality of life refers to the overall well-being of individuals in multiple aspects of life; however, its definition has evolved through time (2). It is a multifaceted concept that comprises perceived psychological, social, emotional, and physical functioning of an individual and is often used to examine the well-being of and burdens in families with neurodevelopmental disorders. WHO defines QoL as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (3). QoL is therefore affected by physical health, psychological state, personal beliefs, social relationships, and relationship with the environment of a person (4).

Quality of life is a difficult concept to define and measure, and there are over a thousand instruments designed to measure the QoL (5). The WHO Quality of Life (WHOQOL)-BREF instrument was developed by the WHO as a result of a global project based on a cross-culturally sensitive concept and is considered appropriate for use across different nationalities (6–8). WHOQoL-BREF measures four domains of QoL, physical health, psychological health, social relationships, and environment, through a set of 24 items (2). In addition to cross-culture applicability, this tool has good to excellent reliability and validity (6–10).

Understanding the different aspects of QoL is essential for parents having a child with developmental disabilities to guide the healthcare workers and policymakers to have a better insight into the struggle faced by parents for planning and implementing different interventions (11).

COVID-19 pandemic, an unexpected catastrophe, has overwhelmed the capacity of the healthcare systems and affected nearly all facets of QoL for the general population. COVID-19 has major ramifications for global health, including the disability community. It has also caused school closures, limited availability of support services, and income loss (12, 13). Children with severe disabilities are physically more demanding and their parents are expected to experience increased caregiver burden during this unprecedented pandemic.

This study aimed to assess the QoL in parents of children with developmental disabilities when compared to typically developing peers in the four domains, namely physical health, psychological health, social relationships, and environment, and overall QoL during the Covid 19 pandemic. Additionally, we aimed to compare the WHOQoL-BREF scores between different developmental disabilities.

MATERIALS AND METHODS

This analytical cross-sectional study with a comparison group was performed at the Pediatric Outpatient Department at the Aga Khan University Hospital, Karachi, Pakistan, after approval by the Institutional Ethics Review Committee (ERC-2020-5476-14718). Parents of children (2–18 years) with developmental disabilities, such as cerebral palsy (CP), autism spectrum disorder (ASD), global developmental delay (GDD), intellectual disability (ID), attention deficit hyperactivity disorder (ADHD), speech delays, learning disabilities (LD), or any syndrome or condition with any of these conditions and receiving services, were included in the study group.

Parents of typically developing children (aged 2–18 years) visiting the Pediatric Outpatient Department (OPD) for routine check-ups or common pediatric problems without any underlying disability were included in the comparison group.

Parents were excluded if they did not understand Urdu or could not fill out the questionnaire.

Parents were included using a consecutive sampling technique. GPower software was utilized to calculate the sample size using the difference in parental QoL of children with disabilities and those of children without disabilities. Literature reports the effective size of the difference in QoL among the two groups is in the range of 0.3–0.4 in various domains (14). Being a ow-middle-income country (LMIC), we anticipated higher intensity of the effect in our population. A sample size of minimum of 100 children per group was required at 80% power and Cronbach's alpha of 0.05 to detect the difference of at least 0.7 points with SD of 2.2; however, we inflated our sample size to 150 participants in each group.

Study participants were administered a questionnaire that contained demographic questions followed by WHO-QOL-BREF-Urdu. The demographic information included the parental age, parent gender, family status(nuclear/joint), marital status (married/divorced/separated/widowed), education level of both parents (no education, can read or write, primary (Grade 5), secondary (Grade 10), graduate, postgraduate), work status of both parents (full-time, part-time, unemployed), monthly family income (<PKRs25K, PKRs25–50K, PKRs50–100K, > PKRs100K), the health status of the parents (healthy/known comorbidity), and age, gender, and diagnosis of the children.

The forms were collected by one of the co-investigators, and the participants were assigned unique codes to protect their identity and to maintain confidentiality.

World Health Organization Quality of Life-BREF questionnaire contained 24 items covering four domains: physical health, psychological health, social relationships, and environment (10). The mean score of items within each domain was used to calculate the mean domain score, after which the mean scores were multiplied by four to make domain scores comparable with the scores used in the WHOQOL-100, as per the instructions of the measure used. Domain scores were scaled in a positive direction with a score range of 0–100 with higher scores denoting higher QoL.

Data Analysis

Data were analyzed using IBM SPSS version 23. The sociodemographic variables were analyzed using descriptive statistics. The Chi-square test and the Mann–Whitney *U*-tests were carried out to observe the statistical differences between the sociodemographic variables of the parents of children with disabilities and those of children without disabilities. An independent samples *t*-test was used to analyze the difference between the scores of parents of typically developing children and parents of children with disabilities on the subdomains of WHOQoL-BREF. A multiple regression was carried out to evaluate which sociodemographic variables can significantly predict the scores of the subdomains and the overall score of WHOQoL-BREF.

RESULTS

A total of 301 participants were recruited for the study, out of which 151 participants were the parents of children without disabilities and 150 participants were the parents of children with developmental disabilities. The demographic data of the participants are given in **Table 1**.

TABLE 1 | Sociodemographic data of parents with children without developmental disabilities (group A) and children with developmental disabilities (group B).

	Group A *n* (%)	Group B *n* (%)
Total no. of parents	151 (50.2)	150 (49.8)
Age		
15–25 years	15 (9.93)	6 (4.0)
26–30 years	53 (35.10)	56 (37.33)
31–35 years	52 (34.44)	56 (37.33)
36–45 years	20 (13.25)	26 (17.33)
more than 45 years	11 (7.28)	6 (4.0)
Gender		
Male	49 (32.45)	60 (40.00)
Female	102 (67.55)	89 (58.33)
Marital status		
Married	149 (98.68)	149 (99.33)
Divorced	0	1 (0.67)
Widowed	0	0
Family status		
Nuclear	60 (39.74)	73 (48.76)
Joint	91 (61.26)	77 (51.33)
Mother's educational status		
No education	3 (1.99)	5 (3.33)
Can read or write	7 (4.64)	6 (4.00)
Primary	11 (7.28)	9 (6.00)
Secondary	38 (25.17)	28 (18.67)
Graduate	68 (45.03)	79 (52.67)
Post-graduate	24 (15.89)	23 (15.33)
Father's educational status		
No education	0	3 (2.00)
Can read or write	2 (1.32)	4 (2.67)
Primary	7 (4.64)	9 (6.00)
Secondary	31 (20.53)	31 (20.67)
Graduate	67 (44.37)	60 (40.00)
Post-graduate	41 (27.15)	43 (28.67)
Mother's work status		
Full-time	18 (11.92)	26 (17.33)
Part-time	7 (4.64)	7 (4.67)
Unemployed/stay at home	125 (82.78)	117 (78)
Father's work status		
Full-time	136 (90.07)	135 (90.00)
Part-time	9 (5.96)	12 (8.00)
Unemployed/stay-at-home	4 (2.56)	1 (0.67)
Monthly family income, PKR		
<25,000	9 (5.96)	8 (5.33)
26,000–50,000	30 (19.87)	40 (26.67)
50,000–s100,000	60 (39.74)	56 (37.33)
>100,000	52 (34.44)	46 (30.67)
Mother's health status		
Healthy	137 (90.73)	144 (96.00)
Known comorbid	14 (9.27)	6 (4.00)
Father's health status		
Healthy	140 (92.72)	136 (90.67)
Known comorbid	9 (5.96)	14 (9.33)

Missing data: Group A missing: marital status: 2; father education status: 3; work status of mother: 1; work status of father: 2; health status of father: 2; gender of child: 1, Group B missing gender: 11; work status of father: 2. Total missing: type: 2; diagnosis of child: 2.

TABLE 2 | Demographic table of parents with children without developmental disabilities (group A) and children with developmental disabilities (group B).

Variable	Group A *n* (%)	Group B *n* (%)
Age of the parent		
<5 years	104 (68.9)	96 (64.9)
5–<12 years	43 (28.5)	45 (30.4)
12–18 years	4 (2.6)	7 (4.7)
Diagnosis		
Autism spectrum disorder	0 (0)	74 (50.0)
Attention deficit hyperactivity disorder	0 (0)	9 (6.1)
Cerebral palsy	0 (0)	23 (15.5)
Global developmental delay	0 (0)	27 (18.2)
Learning disability	0 (0)	3 (2.1)
Speech delay	0 (0)	12 (8.1)

TABLE 3 | Comparison of parents with children without developmental disabilities (group A) and children with developmental disabilities (group B).

Variable	Value	*p**
Gender	2.247	0.134
Education level of mother	3.034	0.695
Education level of father	4.437	0.488
Work status of mother	1.462	0.481
Work status of father	1.985	0.371
Marital status	1.010	0.315

*2-tailed level of significance: $p < 0.05$.

Children with different disabilities were identified. The common disabilities observed were ASD, GDD, and CP. The details are depicted in **Table 2**.

The Chi-square test was carried out to see the statistical differences between parents of children without disabilities (group A) and parents of children with disabilities (group B) in terms of gender, education level, work status, and marital status. No significant differences were observed (**Table 3**).

The Mann–Whitney U-test was carried out between parents of non-disabled children (group A) and parents of children with disabilities children (group B). No significant differences were observed between the scores of the two groups.

An independent samples T-test was used to analyze the difference between the scores of parents of children without disabilities and parents of children with disabilities. Parents of children with disabilities scored *lower* overall when different domains of QoL were considered (physical health, psychological health, social relationships, and environment) (**Tables 4** and **5**). Significant differences were observed for physical health in parents of children without disabilities ($M = 66.09$, $SD = 12.629$) and children with disabilities ($M = 61.80$, $SD = 15.652$); $t\ (285.945) = 2.609$, $p = 0.010$. Similarly, significant differences were also observed for social relationships in parents of children without disabilities ($M = 73.40$, $SD = 16.272$) and those of children with disabilities ($M = 68.37$, $SD = 18.368$); $t\ (297) = 2.509$, $p = 0.013$.

TABLE 4 | Comparison between the mean scores of parents of children without developmental disabilities (group A) and children with developmental disabilities (group B).

Item	*n*	Mean rank	Z	*p**
Age of the parent			−0.812	0.417
Group A	151	146.18		
Group B	148	153.90		
Monthly family income			−929	0.353
Group A	151	154.35		
Group B	148	145.56		
Age of the child			−0.828	0.408
Group A	151	146.63		
Group B	148	153.44		

2-tailed. Level of significance: $p < 0.05$.

TABLE 5 | Comparison of quality of life (QoL) between parents of children without developmental disabilities (group A) and children with disabilities (group B).

Variable	*n*	Mean	SD	T	*p**
Domain 1: physical health				2.614	0.009
Group A	151	66.09	12.629		
Group B	148	61.80	15.652		
Domain 2: psychological health				1.333	0.183
Group A	151	65.51	14.834		
Group B	148	63.25	14.475		
Domain 3: social relationships				2.506	0.013
Group A	151	73.40	16.272		
Group B	148	68.37	18.368		
Domain 4: environment				2.696	0.007
Group A	151	71.99	13.510		
Group B	148	67.79	13.401		
WHO QOL (BREF)				2.96	0.003
	151	89.75	10.35		
	148	85.81	12.56		

2-tailed. Level of significance: $p < 0.05$. SD, standard deviation.

Significant differences were observed for environment between parents of children without disabilities (M = 71.99, SD = 13.510) and those of children with disabilities (M = 67.79, SD = 13.401); t (296.957) = 2.696, p = 0.007. No significant difference was observed in psychological health between parents of children without disabilities (M = 65.51, SD = 14.834) and those of children with disabilities (M = 63.25, SD = 14.475); t (286.994) = 1.333, p = 0.183. The overall score of WHOQoL-BREF also showed a significant difference between children without disabilities (M = 89.75, SD = 10.35) and children with disabilities (M = 85.81, SD = 12.56); t (284.41) = 2.96, p = 0.003.

A multiple regression analysis was carried out to identify potential risk factors for QoL in parents. It was done to predict the score of physical health of parents from the domains of diagnosis of education status, health status, work status, gender of parents, monthly income, family status, gender, age, and diagnosis of children (**Table 6**). These variables statistically significantly predicted the score on the physical health domain of WHOQoL-BREF (14, 276) = 1.886, p = 0.028, with an R^2 of.087. However, the health status variable of the father (B = −8.649, p = 0.016) contributed statistically significantly to the prediction.

A multiple regression analysis was also carried out to predict the score of psychological health of parents from the domains of education status, health status, work status, gender of parents, monthly income, family status, gender, age, and diagnosis of children (**Table 7**). These variables showed statistically significantly predicted the score on the psychological health domain of WHOQoL, F (14, 276) = 2.890, p = 0.000, with an R^2 of.128. However, the health status variable of the father (B = −15.787, p = 0.000) contributed statistically significantly to the prediction.

A multiple regression analysis was carried out to predict the score of social relationships of parents from the domains of education status, health status, work status, gender of parents, monthly income, family status, gender, age, and diagnosis of children (**Table 8**). These variables statistically significantly predicted the score on the social relationships domain of WHOQoL, F (14, 276) = 1.835, p = 0.034, with an R^2 of.085.

However, no single variable contributed significantly to the prediction of the model.

Another multiple regression analysis was carried out to predict the score of the environment on parents from the domains of education status, health status, work status, gender of parents, monthly income, family status, gender, age, and diagnosis of children (**Table 9**). These variables statistically significantly predicted the score on the environment domain of WHOQoL, s (14, 276) $= 4.654$, $p = 0.000$, except for the age of parents variable ($p = 0.044$), with an R^2 of.185. However, the education status of the mother (B $= 2.495$, $p = 0.005$), work status of the mother (B $= 2.680$, $p = 0.027$), and monthly income (B $= 2.833$, $p = 0.003$) variables contributed statistically significantly to the prediction.

A multiple regression was carried out to predict the overall general score of parents on WHOQoL from the domains of education status, health status, work status, gender of parents, monthly income, family status, gender, age, and diagnosis of children (**Table 10**). These variables statistically significantly predicted the overall score of WHOQoL, F (14, 276) $= 3.535$, $p = 0.000$, with an R^2 of.152. However, the education status of the mother variable (B $= 1.727$, $p = 0.027$) contributed statistically significantly to the prediction.

Summary of Key Results

- No statistically significant difference was observed between parents of children without disabilities (group A) and parents of children with developmental disabilities (group B) in terms of gender, education level, work status, and marital status.
- No significant difference was observed between parents of children without disabilities (group A) and parents of children with developmental disabilities (group B) in terms of parental age, monthly income, and age of children.
- Parents of children with disabilities had a lower overall score when different domains of QoL were considered (i.e., physical health, psychological health, social relationships, environment, and overall QoL). Significant differences were observed for physical health, social relationships, environment, and overall QoL. No significant difference was observed for psychological health.

Multiple regression analysis results:

- The health status of the father provided a statistically significant score to the predictive model on the physical health subdomain of WHOQoL-BREF.
- To predict the score of psychological health of parents, the health status of the father added statistical significance to the prediction.
- To predict the score of social relationships of parents, no single variable added statistical significance to the prediction.
- To predict the score of the environment of parents, the education status of the mother, the work status of the mother, and the monthly income added statistical significance to the prediction.
- To predict the overall general score of parents on WHOQoL, the education status of the mother significantly predicted the score on WHOQoL.

TABLE 6 | Summary of a multiple regression analysis of physical health in WHOQoL-BREF.

	Physical health		
	B	**SE B**	**B**
Constant	83.113	16.870	
Type	−8.953	4.911	−0.311
Age	0.623	1.010	0.043
Gender	−0.401	1.795	−0.013
Family status	1.024	1.780	0.035
Education status of mother	1.252	0.996	0.099
Education status of father	0.558	1.095	0.040
Work status of mother	1.279	1.357	0.062
Work status of father	−3.472	2.693	−0.082
Monthly income	−0.820	1.062	−0.050
Health status of mother	0.610	3.684	0.010
Health status of father	−8.649	3.584	−0.159
Age of children	−0.587	1.761	−0.023
Gender of child	0.022	1.735	0.001
Diagnosis of child	−0.770	0.709	−0.184
R-squared	0.087		
Adjusted R- square	0.041		
F for change in R square	1.886*		
No. of observations	290		

**Level of significance: $p < 0.05$. SE, standard error.*

DISCUSSION

Global estimates demonstrate that 15% of the world population is affected by some form of disability (15). Among these, between 110 and 190 million individuals have significant functional limitations and participation restrictions (16). No estimates based on actual measurement of the number of children with disabilities are available; however, a UK study estimated the national prevalence of childhood disability at 7.3% of the population with the highest prevalence of childhood disability seen in the poorest income quintile (17). The epidemiology of disability in Pakistan is limited; however, the prevalence of childhood disability was found to be 5.5 out of 1,000 in rural Sindh (18). According to UNICEF, in developing countries like Pakistan, children at an early age are exposed to harsh living conditions, such as poor sanitation, malnutrition, communicable diseases, and lack of integrated management of childhood illnesses (19).

Children with developmental disabilities often have complex health issues and high unmet health needs (20). As a result of caregiving responsibilities, the parents of children with developmental disabilities report lower QoL and a sense of isolation (21). Children are dependent on parents, thus, having a child with a disability has a negative impact on the parental life, as it requires many adjustments for the family members (22).

This study showed significantly lower scores in the overall QoL compared to control groups. In the four domains of QoL, significant differences were observed in the physical

TABLE 7 | Summary of a multiple regression analysis of psychological health in WHOQoL-BREF.

	Psychological health		
	B	**SE B**	**B**
Constant	84.921	16.539	
Type	−2.192	4.815	−0.076
Age	1.388	0.990	0.095
Gender	−1.775	1.760	−0.059
Family status	0.360	1.745	0.012
Education status of mother	1.214	0.976	0.095
Education status of father	−0.156	1.074	−0.011
Work status of mother	0.299	1.330	0.015
Work status of father	−0.426	2.640	−0.010
Monthly income	−0.955	1.041	−0.059
Health status of mother	−3.008	3.611	−0.052
Health status of father	−15.787	3.514	−0.289
Age of children	−2.708	1.727	−0.104
Gender of child	2.263	1.701	0.078
Diagnosis of child	−0.015	0.695	−0.004
R-squared	0.128		
Adjusted R- square	0.084		
F for change in R square	2.890*		
No. of observations	290		

Level of significance: $p < 0.05$. SE, standard error.

TABLE 8 | Summary of a multiple regression analysis of social relationships in WHOQoL-BREF.

	Social relationships		
	B	**SE B**	**β**
Constant	59.603	20.571	
Type	3.741	5.989	0.107
Age	−1.757	1.232	−0.099
Gender	2.688	2.189	0.074
Family status	0.725	2.170	0.021
Education status of mother	−0.088	1.214	−0.006
Education status of father	0.076	1.335	0.004
Work status of mother	1.940	1.654	0.078
Work status of father	−1.131	3.283	−0.022
Monthly income	1.427	1.295	0.072
Health status of mother	−3.909	4.492	−0.055
Health status of father	−8.390	4.371	−0.127
Age of children	1.961	2.148	0.062
Gender of child	0.107	2.115	−0.003
Diagnosis of child	1.287	0.865	0.253
R-squared	0.085		
Adjusted R- square	0.039		
F for change in R square	1.835*		
No. of observations	290		

Level of significance: $p < 0.05$. SE, standard error.

and environmental domains while no statistically significant differences were observed in the social and psychological domains. While the overall QoL scores were low in all domains for study and control groups, the scores were lower in the study group ($p = 0.01$). These findings are similar to a recent study by Pecor et al. (23) who reported overall lower scores in parental QoL during the COVID-19 outbreak. The lower overall QoL in parents of children with disabilities during the COVID-19 pandemic can be attributed to increased caregiver burden. Another recent study by Shah *et al.* reported worsening of symptoms in pediatric patients with ADHD and an increase in the number of negative interactions with parents during COVID-19 (24).

The physical domain of QoL relates to physical health, sleep, pain, and coping with everyday life and daily physical activities. The mean scores in the physical domain of QoL scores were low overall; however, statistically significant differences were observed in the physical health domain of QoL between the study and control groups. The lower QoL score in the physical domain of QoL is aligned with the findings of prior studies (25–28). The overall low scores in the both study and control groups may be attributed to the ongoing COVID-19 pandemic, which has seen its second and third waves in Pakistan (29, 30). Numerous studies have explored the negative effects of the COVID-19 pandemic on the physical, social, and psychological domains of QoL (23, 31–34).

We did not find any statistically significant difference in the psychological domain of QoL between the study and control groups, which is similar to findings reported by Leung et al. (14). Other studies have reported significant differences in the psychological domain of QoL (25–28). Further research may help to uncover the underlying reasons for this observation.

The social domain of QoL is related to satisfaction with personal relationships and support structure. We did not find any statistically significant differences in the social domain of QoL; however, prior studies have found significant impairment in the social domain of QoL (25, 27, 28, 35). This is an interesting finding, with the plausible explanation being the cultural differences that exist in parental experiences toward child disability (36, 37). The absence of differences in the social domain of QoL could be explained by the Muslim values and the South Asian family-oriented structure of Pakistani society, which may serve as a source of informal support in disability. In a study exploring perceptions toward disability, Ow et al. (38) reported that 48% of Chinese mothers having a child with disabilities had at least one formal source of support; however, none of the Muslim mothers reported needing a formal source of support. Mohamed Madi et al. (36) reported that religious beliefs and cultural norms were major factors in the perceptions of mothers towards child disability in Saudi Arabia, another Muslim majority country.

The environmental domain of QoL is related to physical safety and security, home environment, and health and social care. We observed significantly lower mean scores in the environmental domain of QoL in the study group. This is similar to the findings published in several other studies (14, 25, 27, 28). The greater

TABLE 9 | Summary of a multiple regression analysis of the environment in WHOQoL-BREF.

	Environment		
	B	**SE B**	**B**
Constant	35.784	14.960	
Type	−0.739	4.355	−0.027
Age	−1.692	0.869	−0.118
Gender	2.820	1.592	0.100
Family status	0.034	1.578	0.001
Education status of mother	2.495	0.883	0.209
Education status of father	0.200	0.971	0.015
Work status of mother	2.680	1.203	0.139
Work status of father	3.261	2.388	0.081
Monthly income	2833	0.941	0.185
Health status of mother	6.80	3.267	0.012
Health status of father	−4.426	3.178	−0.086
Age of children	1.793	1.562	0.073
Gender of child	1.915	1.538	0.070
Diagnosis of child	0.432	0.629	0.110
R-squared	0.191		
Adjusted R- square	0.150		
F for change in R square	4.654*		
No. of observations	290		

**Level of significance: $p < 0.05$. SE, standard error.*

TABLE 10 | Summary of a multiple regression analysis of overall WHOQoL-BREF.

	Total raw score		
	B	**SE B**	**β**
Constant	75.890	13.147	
Type	−1.654	3.827	−0.071
Age	−0.508	0.787	−0.043
Gender	2.074	1.399	0.086
Family status	0.351	1.387	0.015
Education status of mother	1.727	0.776	0.168
Education status of father	0.957	0.853	0.084
Work status of mother	2.046	1.057	0.123
Work status of father	−1.762	2.098	−0.051
Monthly income	0.461	0.827	0.035
Health status of mother	−1.189	2.871	−0.025
Health status of father	−5.306	2.793	−0.121
Age of children	−0.125	1.373	−0.006
Gender of child	−0.011	1.352	0.000
Diagnosis of child	0.284	0.553	0.084
R-squared	0.152		
Adjusted R- square	0.109		
F for change in R square	3.535*		
No. of observations	290		

**Level of significance: $p < 0.05$. SE, standard error.*

caregiver burden of having a child with developmental disorders, such as limited socio-adaptive functioning, difficult behaviors and limited social skills, inability to understand the condition of the child, difficulty obtaining the correct diagnosis, and stressful experiences with professionals, contributes to lower scores on the environment domain (25, 26, 28, 36). In addition, limited access to support services due to the COVID-19 pandemic and financial difficulties may further worsen the stress burden.

Prior research has identified that parental QoL is also impacted by the type and degree of disability in the child (39, 40). Disability, depending on the type of disability variable, favors parents having a child with a learning disability, followed by parents having a child with a physical disability, parents having a child with an intellectual disability, and finally parents having a child with Autism, who have the lowest degree of QoL (39).

The subset analysis based on the type and degree of disability could not be performed in this study, due to a disproportionately large sample of parents having children with ASD in the study group, which is a limitation of this study.

Further exploration of this subject with a larger sample size may reveal differences in the degree to which different developmental disabilities in the children affect parental QoL.

This study showed an overall low score in all domains, namely, physical health, environment, social relationships, and psychological health, for both groups. This might be due to the COVID-19 pandemic, and the results were consistent with many other studies done on QoL during the COVID-19 pandemic (23, 41–44). Future efforts aimed at therapeutic interventions in child disability might benefit from focusing on physical and environmental domains of QoL in addition to strengthening support structures through informal approaches.

CONCLUSION

This study demonstrates statistically significant differences in the WHOQoL-BREF scores in the physical and environmental domains of parents of children with developmental disabilities during the COVID-19 pandemic. No significant differences were observed in the psychological and social domains of QoL between the study and control groups.

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/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Review Committee, Aga Khan University Hospital, Karachi, ERC-2020-5476-14718. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

UA: drafting, data acquisition, interpretation, and revision. VB: design of the work, data analysis, interpretation, and drafting. NA: conception, data acquisition, interpretation, and drafting. SJ: conception, supervision of data acquisition, drafting, critical revision, and final approval. All authors contributed to the article and approved the submitted version.

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Developmental Disability: Families and Functioning in Child and Adolescence

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The WHO's International Classification of Functioning, Disability and Health (ICF) provides an integrated framework for health for everyone. Several aspects of this approach to health allow us to see people's lives in a richer and more holistic manner than has traditionally been the case based on diagnosis alone. These features include the positive language (emphasizing in particular "activity," "participation," and "personal factors"); the interconnections of the parts of this "dynamic system," in which every component can influence every other one; and the formal inclusion of "contextual factors"—personal and environmental—that are otherwise too easy to take for granted and then ignore. This paper addresses the "environmental" dimension of the ICF framework—specifically referring to "family" as the central environmental force in the lives of children and adolescents. The perspectives of the author are those of a developmental pediatrician, whose career has focused on children with conditions that challenge their development, and their families. Lessons learned from a lifetime of work—including teaching and research as well as clinical services—are offered. Particular emphases will be on (i) the importance of focusing on the family in a non-judgmental "family-centered" way; (ii) how conceptual ideas about child (and family) development and parenting are as important as technical approaches to intervention; and (iii) how the ICF framework "allows"—indeed encourages—such a focus to have value and importance equal to the best of biomedical interventions. Examples from current research will illustrate how these ideas can be implemented.

Keywords: childhood disability, ICF, F-words, family well-being, family-centered service

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INTRODUCTION

The Author's Perspective

This paper is not a conventional literature review, but rather a personal essay about a topic in which the author has longstanding interest and engagement. It draws on my perspectives and experiences as a developmental pediatrician—a clinician, teacher, and health services researcher in childhood disability. The ideas offered here are predicated on two statements of the obvious. First, in the field of child health, we work with families. Every child we ever see comes wrapped in a family. By "family" I refer to one or more responsible adults who have a central on-going caregiving role to protect and nurture that child. In the current era, families appear in a variety of constellations. Whatever their make-up, however, they must be the unit of our clinical attention. That means that we cannot consider only the child without an equal interest in, and focus on, their "family." Second, if it is true that "It takes a village to raise a child," it is equally true that a child's challenges of health or development affect the whole family (and beyond, as I shall argue). This means that the child's issues cannot be addressed solely as an individual's predicament.

Modern Thinking About Health

The centrality of the family in children's lives is not simply the author's personal view. The World Health Organization's 2001 framework for health was formulated within its InternationaI Classification of Functioning, Disability and Health (1) (the ICF). This framework formally identifies "contextual" factors that include "environment." The nuclear environment of the child is their family. In its efforts to bring the ICF framework to life, CanChild researchers wrote a whimsical paper about "The F-words for Child Development" (2) (see **Figure 1**), in which the "f-word" family is used to illustrate the "environmental factors" component of the ICF.

What are the implications of this interest in family? I believe that all our considerations of the health, development and well-being of children and youth need to be built around a set of values, namely: (i) an understanding of each specific family, with its strengths, resources and challenges; (ii) acceptance of each family's individuality and culture, within which, as service providers, we need to operate; and (iii) the importance of a working relationship with families based on a shared, mutually respectful partnership. The famous physician and educator Sir William Osler offered the opinion that: "The good physician treats the disease; the great physician treats the patient who has the disease." (3). In our field, one might rephrase this idea and state: "The good child health professional treats the child's condition; the great child health professional treats the family of the child who has the condition."

How Can We Think About Our Work With Families?

In reflecting on how health professionals can work most effectively with families of children and youth with chronic conditions, we will explore both the *processes* by which we should engage with families, including consideration of the values that underpin our interactions, and the *content* and nature of our advice and counsel. Both the processes and content of our work with families continue to evolve, as will be outlined briefly in the respective sections of this paper.

WHAT DO WE MEAN BY THE *PROCESSES* OF WORKING WITH FAMILIES OF CHILDREN AND YOUTH?

Family-Centered Service

The concept of "family-centered care" emerged in the US in the 1960s, promoted by parents of children who required hospitalization, and whose engagement with their child's care in hospital was very limited. In fact, in the later 1960s when I trained at a major children's hospital, there were *visiting hours for parents*! One hopes that in the twenty-first century this strange situation is a historical anomaly, and no longer a reality anywhere.

What do we mean by the term "family-centered service" or FCS? (We prefer the term "service" to "care," as professionals are "service providers.") We see FCS as "…a set of values, attitudes and approaches to service for children with special needs and their families. The family works together with service providers to make informed decisions about the services and supports the child and family receive. In FCS, the strengths and needs of all family members are considered." (4) FCS is built on three premises: (i) parents/caregivers are the experts on their child's needs and abilities; (ii) each family is unique; and (iii) the family is the constant in the child's life.

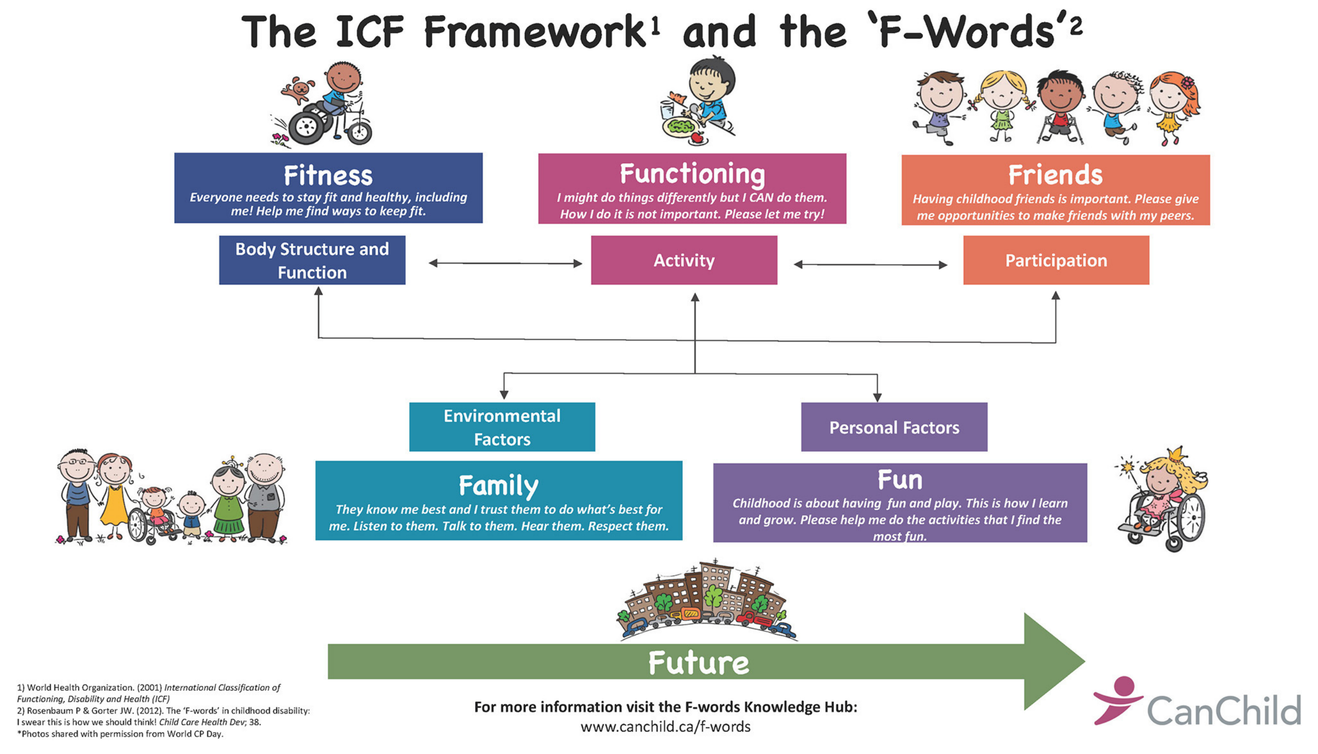
How Does FCS Work? How Do We Do It?

If one accepts the premises outlined above, it becomes apparent that we need to work in a mutually respectful partnership with families. As the world's experts on their child (they know them best), parents/caregivers must be involved in decision-making about issues that concern their child and family, and should have ultimate responsibility for the care of their children. The uniqueness and individuality of every family means that we cannot assume anything about their values or beliefs based on ethnicity, religion, skin color or any other external characteristic. Rather, it is essential to understand their predicament as they experience it. A wise parent expressed this idea very clearly when she told me: "You have textbooks; we have story books!" (5). In other words, to provide the optimal services for children and their families we need each other's special insights—service providers' understanding of the clinical realities of diagnosis, prognosis, effective interventions for these conditions, and so on, and families' stated values, beliefs, hopes, expectations, resources, and so on.

Yes, but What About…?

FCS concepts and practices have taken root in many settings, but there remain people who may be uncomfortable with the idea of FCS. They wonder whether we are now simply expected to do a family's bidding, regardless of our own judgment; or whether we have to wait for parents to decide that they want and need before we can act. They are asking, in effect, "Is the tail wagging the dog?" They also wonder whether FCS actually matters. There are good answers to both these concerns.

Needless to say, we need to understand families' requests and wishes, as a basis for discussion and shared problem-solving. This can happen when we build mutually respectful relationships with families, whereby everything can be discussed and addressed thoughtfully. Our perspectives and interpretations of parents' concerns can be the foundation for conversations about the deeper meaning of a parent's wishes, and possible approaches to address them. As an example, imagine a young child who cannot speak clearly, for whom the parent's main goal is for the child to talk. We can reframe the situation to help parents see that what they want is to understand what's on their child's mind—in which case alternate modes of communication may be an excellent approach while the child continues to develop speech (6).



Everyone needs to stay fit and healthy, including me! Help me find ways to keep fit.

I might do things differently but I CAN do them. How I do it is not important. Please let me try!

Having childhood friends is important. Please give me opportunities to make friends with my peers.

They know me best and I trust them to do what's best for me. Listen to them. Talk to them. Hear them. Respect them.

Childhood is about having fun and play. This is how I learn and grow. Please help me do the activities that I find the most fun.

For more information visit the F-words Knowledge Hub:
www.canchild.ca/f-words

1) World Health Organization. (2001) *International Classification of Functioning, Disability and Health (ICF)*
2) Rosenbaum P & Gorter JW. (2012). The 'F-words' in childhood disability: I swear this is how we should think! *Child Care Health Dev*; 38.
*Photos shared with permission from World CP Day.

FIGURE 1 | The ICF framework and the F-words.

Here is another strategy that can be effective: when parents are uncertain how best to proceed, we can lay out the range of approaches that other families in this predicament have considered. By providing families an understanding of their options, and listening carefully to their questions, we may be able to "precipitate" a course of action for them based on what they ask us, and on their ideas. Using their words, we may be able to say "It sounds to me like following course of action would make most sense to you at this point." For example, parents may be uncertain as to whether their child with toe-walking associated with spasticity in calf muscles should have any intervention, and if so, might this be a brace or botulinum toxin injections? Parents seeking long-term changes may find the bracing option more palatable than repeated injections, whereas parents wanting to know if changes in muscle activity improve gait are more likely to opt for a trial of botulinum toxin. This approach has, on many occasions, helped me in my work with families—an example of how we can help them help us help them!

Another less appreciated aspect of being interested in families is to understand what the grandparents know and think (7, 8). We most often meet "parents"—the people who bring their children to us with their concerns. We can forget that most (young) parents have their own living parents—and are therefore also someone's "child!" The people we are seeing as parents are thus living in a "generational sandwich," and their parents may "suffer twice"—for their grandchild and for their (adult) child's predicament. This reality opens up opportunities for us to ask about and try to understand the roles and influences of grandparents in the lives of the parents and children with whom we are working. Offering parents the opportunity to invite the grandparents to attend clinic visits with them is a powerful and very rewarding way to be family-centered.

It is a statement of the obvious that grandparents are a generation older that their (adult) children. Thus, their perspectives, beliefs, and attitudes about disability may be dated relative to, for example, current ICF-based thinking. Being able both to hear and explore their questions and fears, and share ideas such as those offered here, enables them to at least consider their views in light of concepts and evidence that they otherwise had no reason to know. Although I have never explored this approach systematically, I have done this scores of times and recall these experiences clearly as among the most memorable of my clinical career (I will add that many parents, when offered this opportunity to engage their parents, are convinced that the grandparents will not be interested—only to have the grandparents seize the opportunity and engage actively.)

We have argued that "Parenting is a dance led by the ever-changing child" (9). I believe that this metaphor also applies to our relationships with families. Parents develop and change, become better informed with time and experience, and see their changing child's abilities and needs emerging. When we have been able to create relationships that are built on trust, respect and confidence in each other, each of us can help the other succeed in our shared "dance" toward goals for their child and family.

Yes, but Does FCS Matter?...

As for the value of FCS to families, there is good evidence of important relationships between parents' reports of their experiences of FCS, using validated tools created with parents for parents (10) and parents' reports of their satisfaction with services, their mental health, and the stress they experience in working with us (11).

In summary, FCS can be learned and practiced, and it does make a difference to parents and families. FCS does "work" (12).

IN WHAT WAYS DOES THE *CONTENT* OF OUR ADVICE MATTER TO PARENTS AND FAMILIES?

What Do They Know Already?

It is important to remember that as specialist service providers we rarely if ever are a family's first-contact experience. That means that it is essential to understand what families "know" already. It is on that foundation of facts, beliefs, information, misconceptions, expectations and values that we will build our relationships with families, as outlined above, and add our advice and ideas to the mix of what is already "there."

After introductions, and my routine opening question to all parents—"What do you want to boast about concerning your child?"—we need to understand what they have been told. An open-ended question such as "Tell me about your child" or "Tell me how I can be helpful?" allows parents to tell their story in their own words before we bombard them with questions. We want to listen for and learn: Has there been a diagnosis given? What was the tone of the conversations that preceded the referral to us? What have parents been told we (service providers) can/will do? An additional good way to find out both what parents are focusing on and what they have learned is to ask what websites they have explored.

I am especially interested in the idea of what I call the "catalog of doom"—the long list of things that parents have been told their child will *not* be able to do. Conversations with families in which people present a bleak outlook can be devastating to the parents, and are often ill-informed. They may be offered with good intentions, because of a belief that preparing people for the worse is appropriate. My own approach to this "catalog" catastrophizing is to remind parents of three ideas: first, we are rarely as accurate at predicting as we would wish to be; second, children are constantly changing and developing, so an early effort to understand the "story" and its outcome should be avoided, because the plot will continue to unfold; and third, I tell parents that the children rarely "listen" to these bleak prognostications, and often achieve much more than was originally predicted! [A thoughtful exploration of this complex issue of communicating "bad news" is offered by Siegler (13).]

What Are We Offering in Our Counseling and Advice?

Building on the details that parents have reported in our initial conversations described above, and the specific questions they have for us, we need to address their issues as clearly, specifically, and honestly as possible. Parents may have been told that our services will offer "therapies," with a more or less explicit implication that these will fix their child's issues. Parents have often read about specific therapies—be they chemical (e.g., botulinum toxin injections), physical (e.g., physical, occupational, and speech-language therapies), technical (e.g., bracing or special Velcro outfits), electronic (e.g., muscle stimulation treatment), etc.—that are reported to be exactly what their child needs. Some of this advice may be relevant to their situation, while other elements may reflect advertisements for what are often referred to as "complementary and alternative therapies" of uncertain value (14, 15). At the same time, if we are honest, we have to acknowledge that much of our accepted conventional intervention is also of uncertain value much of the time.

The reality is that we almost always recommend "programs" of intervention with a mix of ideas and advice, and rarely offer just individual "treatments." This is because we are trying to promote functioning and development, and not simply to address a specific impairment as one might do in rehabilitating and strengthening weakened thigh muscles after a broken leg.

What Can We Do Differently?

For these reasons, I believe that we should provide both a context for, and an interpretation of, parents' concerns, and offer a modern view of our thinking. With colleagues in Australia, our CanChild team is involved in a research study to assess the impact of the introduction of a specific programmatic set of ideas we believe are important for parents. These five interactive workshops present (i) WHO's ICF ideas about health and the F-words framework; (ii) Ideas about child, family and sibling development; (iii) "Parenting is a dance led by the child;" (iv) Strategies for looking after myself; and (v) Communicating, collaborating and connection with others (about our child and family). It is hoped that what one parent has called "early intervention for parents" will positively influence their approach to their child and family's development and functioning (16).

The ideas we are promoting are built around the F-words concepts described earlier in this paper. We want to encourage parents to take a *strengths-based approach* to child and family *development*; to help families understand that while we cannot "fix" developmental impairments we can *promote functioning*—however it is done—and enable parents to see their children as *having capability* even in the face of impairments. It is essential to *recognize the enormous diversity in human functioning*, and to move away from the tyranny of "normal" and *celebrate* *achievement, however it is accomplished*. Finally, we encourage people to take a *life-course approach* to their predicaments, based on their ever-changing child (and family) and the continuing evidence of what they can do if given the opportunities.

We are offering these ideas to parents in an effort to help them "reset" their thinking about themselves, their child, their control over their lives. Understanding the concepts of *capacity* and *performance* can be useful, reminding parents that what we do at our best (*capacity*) is not always how we *perform* on a day to day basis. This idea can be important if it allows parents, and us, to explore the factors associated with the gap between capacity and performance, and to recognize opportunities—often with environmental interventions, but also perhaps with counseling—to minimize this gap and maximize performance.

That All Sounds Lovely in Theory, but...

Our work to promote understanding and uptake of the F-words for Child Development is enabling us to acquire a considerable body of evidence of the currency and power of these ideas around the world. For many parents, these ideas are transformative. I can do no better than to end this essay with the words of parents and colleagues, whose anecdotes and testimonials speak volumes about how the *content* of our ideas has impacted them and their families.

> *"Thank you for making [a podcast about an editorial (6)] as this came in perfect timing for our family. X... (child with CP) is working on potty training by herself and the other day (we didn't know she was in the bathroom) she had a BM and tried to clean herself, but obviously it was messy and got all over the toilet. I had to take a deep breath because I wanted to yell at her for the mess, but then you popped into my mind. At one of our check-ups with you, a long time ago, you had said you cared about function rather than it being perfect. So instead of yelling at her I applauded her for the great try and that we would work on getting better." (A parent, May 2021)*
>
> *"Today in clinic I saw a child (who has CP and is functioning at a GMFCS V)—whose mother attended CP-NET Science and Family Day last Wednesday. She reported to me that her entire approach to raising her son has shifted from one of 'fixing' to embracing the F-words and a wellness approach. She came to clinic with her goals related to the F words all worked out and felt very empowered! I couldn't be more pleased." (From a colleague, May 2017)*
>
> *"Today I saw a 6-year-old girl with CP GMFCS V, refugee from XXX via YYY, parents tried Stem cell therapy in ZZZ and came here (Canada) with hopes to help her. We had a long consult with a whole team of therapists, social work, resident and myself... trying to answer all the questions they had. I tried to explain what we understand by a functional approach and what the purpose of therapy is and that the exact etiology (nyd) probably won't change this approach. At the end, I showed them the F-words poster in Arabic, they read carefully, asked if these words are meant to be the child speaking and I confirmed. The mom commented under tears "this is beautiful, that's what I wish for my daughter." (From a colleague, July 2019)*

In summary, our work with families of children and youth with long-term challenges of health or development can be incredibly rewarding and productive. The ICF's biopsychosocial framework for health, and the F-words ideas that bring these concepts to life for parents and families, provide a common language and approach for families and service providers to connect and share ideas. The f-word "family" reminds people that the nuclear environment of the child is potentially the most important of all, and demands attention from us regarding what we do and how we do it. By working as a team, in partnership with parents (and with children whenever possible), we can formulate shared goals, strive to empower families and young people to achieve their goals however they do so, and live lives that are rich and successful in their own terms. What we say and do, and how we do it, are under our control. I hope this paper has provided some evidence to support this assertion.

AUTHOR CONTRIBUTIONS

The author confirms being the sole contributor of this work and has approved it for publication.

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Reconceptualizing the Family to Improve Inclusion in Childhood Disability Research and Practice

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The World Health Organization's International Classification of Functioning, Disability and Health recognizes that environmental factors impact well-being and life participation for children with disabilities. A primary environment in which children grow and learn is the family. The importance of family has long been recognized in family-centered practice and family-centered research. Although family-centered services and research have been critically explored, the concept of family has received less critical attention in rehabilitation literature. The family construct is due for an updated conceptualization with careful consideration of the implications for childhood disability rehabilitation practice and research. Interrogating the family construct asks questions such as: who is included as a part of the family? Which family structures are prioritized and valued? What is the potential harm when some families are ignored or underrepresented in childhood disability practice and research? What implications could a modern rethinking of the concept of family have on the future of childhood rehabilitation practice and research? This perspective article raises these critical questions from the authors' perspectives as parents of children with disabilities, child focused rehabilitation professionals, and researchers that focus on service delivery in children's rehabilitation and family engagement in research. A critical reflection is presented, focused on how the construct of family affects children's rehabilitation practice and research, integrating concepts of equity, inclusion and human rights. Practical suggestions for children's rehabilitation service providers and researchers are provided to aid in inclusive practices, critical reflection, and advocacy.

Keywords: family, childhood disability, rehabilitation, family-centered services, inclusion, family systems theory

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INTRODUCTION

As parents (JL, RM), practitioners (MP, MR), and researchers (MP, MR) we acknowledge that children grow and develop in the context of their family. Family is the most immediate and powerful influence on a child's formative years that affects their lifelong trajectory (1). As such we focus on the family as a part of care and generally recognize family-centered service as a preferred framework to guide service delivery for children with disabilities (2). Family-centered services have been described as a philosophy and framework that recognizes families as the constant in a child's life, values parents' knowledge of their child, and their partnership in services (3). Despite the widespread adoption of family-centered service there are reported challenges with implementation, such as difficulties collaborating between parents and service providers (4) and a lack of services that focus on the well-being of the whole family (5). The language used to refer to family caregivers is complex reflecting the informal and typically unpaid role that many family members assume when caring for someone with a disability (6). The concept of family-centered services has been critically examined, recognizing the tension between the family's therapy preferences, cultural norms and expectations, and competing organizational considerations such as service delivery models and funding (7).

Similar to family-centered services, family-centered research has been proposed as a form of patient-oriented research that prioritizes families' interests and perspectives (8). In family-centered research an equal partnership can be created that invites families to engage throughout the research process and share their ideas and critiques in informing the study design, conduct, and knowledge sharing (8). The concept of patient-oriented research has drawn critical attention, raising questions of how to engage patients authentically (9), how to compensate patients (10), how to engage families that are underrepresented in research (11) and regarding the methods that may be used to promote reflexivity throughout the collaborative research processes (12). The conceptual theory underlying patient engagement in health settings and what it means to include 'the patient voice' has been critically explored (13). Raising these questions has helped to advance practices in this area, generating recommendations that guide researchers to examine and inform their practices to improve quality and inclusivity in research.

While family-centered service and family-centered research have been critically examined, the underlying concept of *family* has yet to undergo formal critical scrutiny as applied in childhood disability research and practice. Traditionally childhood disability scholars and practitioners focused on the child and their health condition in isolation (14). However, application of a biopsychosocial model in the World Health Organization's International Classification of Functioning, Disability and Health (ICF) shifted thinking to a contextual view of the child, recognizing that children's function is impacted by both children's health conditions and their social environments (e.g., families, communities) (15). Sociologists and psychologists have a history of studying the family unit in which family systems theory, social ecological models, and structural-functional theories were used to examine the interdependence of family member identities, roles, and functions (14). These micro and macro-level theories account for how family members construct their individual and family identities as situated within their culture.

Traditionally, the nuclear family structure that represents white heterosexual norms and values was viewed as the typical or even ideal family, including a married mother and father residing with their unmarried biological children (14). Overtime, cultural and legal norms have expanded to recognize other family structures that include adoptive families, divorced families, and step-parent relationships (16). In some cultures, multigenerational families are recognized, with grandparents holding prominent roles in the child's care and home responsibilities (14). There continues to be controversy over whether lesbian, gay, bisexual, transgender (LGBT) couples are recognized by law and allowed to marry, and these families may face additional prejudice and judgement in their communities (17). Little is known about the experiences of LGBT parents who are raising children with disabilities (11, 14). Clinical practice and research with foster-families are complicated by informal kinship arrangements, formalized kinship, or non-kinship foster family status (18). There are family structures that are rarely included in research, such as polyamorous and polygamous families that may face negative judgement and marginalization when seeking health care for their families (17, 19).

It has been said that "it takes a village" to raise a child and this may be especially true for children with disabilities and their families; however "the village" is typically unrecognized in rehabilitation services and research (20). A family's culture and ethnicity may shape "their village" to include cousins, aunts, uncles, grandparents and non-relative members, such as Godparents and these roles may reflect culturally formed expectations regarding financial support, caregiving, and provision of advice (14, 17, 21). People with disabilities (22) and parents of children with disabilities (21) have also included friends and peer support networks in "their village." These individuals contribute to their well-being, due to an empathetic understanding of peoples' needs, and the availability and willingness to provide physical, emotional, spiritual care, or guidance (21, 22). Despite the high value placed on these relationships and their potentially transactional nature, friendships are not legally recognized with the same rights as family relationships (22).

Family Systems Theory is applied in this article to conceptualize family as:

i) a system of individuals that are bonded together through their co-constructed identity as family members,
ii) people with roles and functions that tie members to one another and influence individual and collective family outcomes,
iii) the sharing of a social location in a broader environment that (a) shapes families' identity and (b) allows families to shape the culture in which they are embedded.

This article provides a critical reflection on how the conceptualization of family affects rehabilitation practice and research for children with disabilities and their families. The parents on our authorship team (RM, JL) initiated conversation with the researchers (MP, MR) to raise concerns about how family is defined in the childhood disability research and care contexts. They described the high demands placed on mothers, devaluing of non-related family members in their social support networks, and the need to consider research and policy implications (e.g., who is counted on research demographic forms and who qualifies for respite care). We advanced these ideas and generated recommendations through iterative discussions and draft revisions that integrated theoretical concepts and literature with examples from parents', practitioners', and researchers' lived experience.

HOW DOES THE CONCEPTUALIZATION OF FAMILY AFFECT CHILDREN'S REHABILITATION PRACTICE?

The definition of family used in a clinical setting has major implications for service delivery. At the outset of service, legal guardians, or parents need to consent to a referral, assessment or therapy plan (23, 24). They often provide insight on goals for the child and take responsibility for the implementation of home programs. Parents typically have the right to access information about the child's therapy and progress, for example through the receipt of written or verbal reports from service providers or electronic health records (24). One family member is often called upon to share their child's therapy information with other people who are involved in the child's life, for example their partner, daycare providers, grandparents, or other professionals (25). Often tasks such as providing consent, setting goals, sharing information, and implementing home programs are taken up by the parent who attends therapy, even in dual parent families (25, 26). The heightened demands placed on a parent in single parent families or families that co-parent when only one parent attends therapy have been reported and should be considered when developing a service plan with parents (26).

How Can We Create Clinical Environments That Are Inclusive of Diverse Family Structures?

1. At the point of intake ask open ended questions to determine how the family members view their family. For example, "Would you mind telling me about your family?" This may provide insightful information about the adults in the child's life, siblings, step-family members, and living situation. Use these insights when completing contact information forms and case history questions.
2. Consider sharing your pronouns to signify that clients and family members are invited to do the same. Consistently use the pronouns that people tell you they identify with when you interact with family members and in clinical reporting. Use gender inclusive language when referring to family members, for example, "does your partner also work during the day, what is their phone number?"
3. Ask families about who they would like to be a part of their therapy and how they would like to communicate with you. For example, a grandparent may work with you because the child is cared for by the grandparents during the day. Can that be accommodated? Would parents like for you to send them progress updates directly or via the grandparents?
4. Use available literature and conversation with clients to reflect on your own biases and heighten your understanding of the care experiences for people whose family structures do not match dominant cultural ideas of family. For example, would individual therapy be more comfortable for a transgender parent than a group program where they may fear and experience judgement from other families?

What Are the Potential Risks When a Nuclear Family Structure Is Reinforced in Existing Rehabilitation Practices?

If children's rehabilitation service providers do not think critically about how diverse family construction affects their practice, we risk reinforcing existing stereotypes and barriers to service use. Families may feel unwelcome and avoid or delay service use. This may lead to missed opportunities to provide early intervention for children with a cascade of negative outcomes (e.g., missed diagnosis, delayed therapy). In assessment, a holistic understanding of the child's skills, needs, and goals may be lacking if only one parent provides information and other people who are close to the child are not invited to participate. In therapy, service providers who do not discuss family member roles may make erroneous assumptions about the resources and supports that are available to facilitate therapy participation (e.g., bringing children to appointments or doing home practice). Often the burden for sharing information about therapy progress and plans is carried by the adult who attends sessions. A fulsome understanding of who is regarded as family and obtaining necessary consents may allow the service provider to directly share relevant information with each individual. This would reduce the responsibilities for the adult who attends appointments and potentially avoid conflict when sensitive information or recommendations need to be communicated (e.g., a diagnosis).

HOW DOES THE CONCEPTUALIZATION OF FAMILY AFFECT CHILDREN'S REHABILITATION RESEARCH?

The conceptual definition of "family" affects who is recruited, the data collected, and the findings produced in family focused research (16). This is particularly true for families that do not fit traditionally recognized structures, such as, families created through surrogacy or adoption, divorced or blended families, LGBT families, polygamous families, or multigenerational families (16, 17). Researchers who focus solely on the nuclear family may miss opportunities to understand and appreciate broader conceptualization of families that also includes social support networks (e.g., religious communities) and kin who may be relatives or non-relatives (14, 16, 21).

When designing a study protocol, developing inclusion-exclusion criteria, and creating recruitment materials it is necessary to carefully consider the desired sample and to justify the accompanying methodological choices (e.g., sampling strategy, recruitment terminology, venues, and processes). These decisions have tangible implications for the research completed and the potential application of findings. For example, stress and coping in families of children with disabilities are frequently studied, however close examination of this literature demonstrates that it is typically mothers' stress and coping that are documented, with few studies on siblings, grandparents, or fathers (20, 27). When family researchers attempt to include diverse members, such as stepparents, they may face barriers due the stigma associated with particular labels or a presumption that general labels (e.g., sibling) do not include step-siblings or half-siblings (16). In polygamous families, individuals may use invented language, such as "tribal aunt" to signify belonging, and these terms may be unknown and underutilized by researchers (17). By intentionally or unintentionally excluding the people who consider themselves to be family members of a child with a disability, we are missing opportunities to generate data that would inform our understanding of their perspectives and to inform clinical practices.

Research rarely examines the experience of children with disabilities who are raised by parents who are LGBT (11, 17) and siblings and grandparents are often overlooked in the literature. Siblings can be highly involved in therapy and may assume care responsibilities as adults, however research focuses more on parents therapy involvement (28). Research about grandparents of children with disabilities has increased over the past decade, revealing heterogeneity in grandparents' acceptance of the disability, frequent worries for family, high levels of support, and family cohesiveness (29). A paucity of research may reinforce exclusion, under representation and stigma for diverse families, for example families who have undergone divorce or remarriage (16) or polygamous families (17).

Data analysis provides researchers with an opportunity to critique their assumptions, for example are you comparing the family experiences and outcomes to a presumed ideal or normative family type? Are you applying a deficit-based lens to problematize unfamiliar family experiences and could this be reframed from a strengths-based position? For example, research on polygamous families indicates that these families may have challenges and fears about disclosing their relationships to their children and about child custody. However, research also notes the benefits of collaborative parenting in polygamous communities such as, shared resources and increased adults to spend time with the children (17). Engaging members of the community when creating your research question and during data analysis may help to ensure that these strengths-based questions and interpretations are not overlooked (11).

How Can We Create Research That Is Inclusive of Diverse Family Structures?

1. When designing a study, carefully justify your participant selection criteria and choose language that matches the language used in your target communities. This may require collaboration or pilot testing of your recruitment materials with members of the chosen community.
2. If your research is about families of children with disabilities consider whether your question is inclusive to all family members (e.g., siblings, grandparents) and family structures (e.g., kinship communities, co-parents who do not live together) and justify your decisions. Check whether your data collection forms (e.g., demographic questions) and survey or interview questions allow all family members to contribute data and be included (e.g., how are gender questions worded)?
3. Embed critical reflexivity into your research to interrogate your own position and beliefs and the potential impact on your research. When it is appropriate invite collaboration and critical questioning from people who have a family experience that is different from your own.

What Are the Potential Risks When a Nuclear Family Structure Guides the Research?

If researchers do not embrace a holistic definition of family that is inclusive of the people recognized as family in the lives of children with disabilities, there is a risk of excluding people from research and reinforcing a narrow understanding of family life. This limited evidence-base will make it challenging to draw from the study findings for use in clinical practice with individuals beyond the client and their mother and father. There will be missed opportunities to understand and reinforce the value and strength in diverse families.

DISCUSSION

The discussion and recommendations presented thus far were intended to support children's rehabilitation service providers and researchers to (i) develop inclusive practices and (ii) consider the potential risks of maintaining focus on the nuclear family. While we hope that these strategies may be taken up to improve research and practice at the individual level, we recognize that collective advocacy is needed to promote widespread acceptance of diverse families of children with disabilities.

The WHO-ICF highlights three environmental factors that are relevant to this discussion of family: support and relationships, attitudes, and services, systems, and policies (30). Under supports and relationships there is clear evidence to promote the inclusion of families in service delivery and research; however, advocacy may be needed to expand consideration of "who counts" as a family member. For example, do regulatory bodies and privacy guidelines allow grandparents who hold informal guardianship roles to consent to therapy on behalf of a child who resides with them, even if parents hold legal custody? Our clinical experiences as Speech-Language Pathologists and Occupational Therapists in Ontario, Canada suggest that parental consent is required for all treatment decisions, unless legal guardianship has been transferred. Perhaps advocacy is needed to allow flexibility in these circumstances, such that parents could provide a blanket consent allowing grandparents to make therapy related decisions. In research, manuscript reporting guidelines may request that authors justify the congruency between their research question and sample. For example, if your question is about well-being in *parents* of children with disabilities were both mothers and fathers recruited? If it was about *family* well-being, were siblings, grandparents or other family members included? Grant priority funding may be allocated for groups that are often excluded in childhood disability family research, e.g., informal kinship or friendship networks, LGBT parents, polyamorous and polygamous families.

Attitudinal environmental barriers to functioning indicate that children with disabilities and their families are likely to experience disability related stigma and this experience may be heightened for families who hold other identities that are devalued in society (e.g., LGBT parents, families with low socioeconomic status, and racial or ethnic minority families) (14). Implicit bias training for children's rehabilitation service providers and researchers may help people to increase awareness of their own biases about families and to mitigate the potential for negative consequences in client care (31) and research conduct (e.g., how questions are framed and data is interpreted).

Service providers and researchers have a critical role to play in advocating for health services, systems, and policies that promote the inclusion, functioning and well-being of children with disabilities and their families (32). At the broadest level, we should align with Article 5 of the United Nations Convention on the Rights of the Child (CRC), which recognizes that the following people have a duty help children to exercise their own rights over time "…parents or, where applicable, the members of extended family or community as provided for by local custom, legal guardians or other persons legally responsible…" (33) and Article 2, which protects children from discrimination, including that which stems from their parent's or guardian's sex, ethnic or social origin, political or other opinion (33). The WHO-ICF personal factors may aid researchers and service providers in identifying aspects of the individual's background (e.g., age, race, gender) that may interact with a health condition and environment to impact function and participation in everyday life (34). These applications of the CRC and ICF may support service providers' and researchers' efforts to critically examine the identity of clients and families and equitably support their inclusion in services and research.

We recommend that future research be conducted with families, clinicians and researchers to: (i) understand how they conceptualize family, (ii) identify biases in how families experience inclusion in care and research, (iii) promote critical reflection in practice and research, and (iv) advance inclusive practices with diverse families in clinical care and research. To honor the CRC and enact family-centered care and research the ICF personal and environmental factors can be usefully applied to critically examine our conceptualization of children and their families and advocate for full inclusion in rehabilitation services, research, and society.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study reflect the personal experiences and reflections of the authors, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

MP was primarily responsible for writing this article. The ideas presented were developed by all authors from their combination of lived experience as parents of children with disabilities, clinical experience in children's rehabilitation, and academic research. All authors have had an opportunity to review, revise, and approve the article. Authors share responsibility for the information presented in this article.

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PERSPECTIVE
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Critically Examining the Person–Environment Relationship and Implications of Intersectionality for Participation in Children's Rehabilitation Services

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Participation of children in rehabilitation services is associated with positive functional and developmental outcomes for children with disabilities. Participation in therapy is at risk when the personal and environmental contexts of a child create barriers to accessing services. The International Classification of Functioning, Disability and Health (ICF) provides a framework for conceptualizing the personal and environmental factors linked to a child. However, it does not facilitate critical examination of the person–environment relationship and its impact on participation in children's rehabilitation. This perspective study proposes the use of intersectionality theory as a critical framework in complement with the ICF to examine the impact of systemic inequities on the participation in therapy for children with disabilities. Clinicians are called to be critical allies working alongside children and families to advocate for inclusive participation in children's rehabilitation by identifying and transforming systemic inequities in service delivery.

Keywords: ICF, intersectionality theory, participation, personal factors, environmental factors, children's rehabilitation, childhood disability

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INTRODUCTION

The International Classification of Functioning, Disability and Health (ICF) holistically conceptualizes everyday functioning and disability across the interconnected domains of the following: body functions and structure, activity, and participation (1). The ICF defines participation as the "involvement in a life situation" (1, 2). Participation at home, school, and community has positive outcomes for child development and provides children the opportunity to develop skills required to support the transition into adulthood (3–6). Children with disabilities experience opportunity limitations and restrictions in participation when compared with their peers without disabilities (7–9). Given its association with improved developmental outcomes, understanding, measuring, and optimizing participation for children with disabilities is a common aim in children's rehabilitation (3, 6, 10–12). The ICF provides a common language to classify, understand, and study health and its related outcomes (1). Although the ICF can be used as a tool to raise awareness of necessary social change (1), it does not facilitate critical examination of how systemic health inequities are sustained by the uneven distribution of power and resources as well as dominant social practices (13, 14). Application of a critical lens could enhance the potential of the ICF to advance social change through the critical examination of the personal and environmental factors that can impact participation and health outcomes.

For children with disabilities, participation in rehabilitation is linked with positive functional outcomes (15). Participation in children's rehabilitation can be described as the active involvement of children and parents in all aspects of the therapeutic process (16). Goals related to optimizing life participation for children with disabilities can be a focus of rehabilitation services (17, 18). Given the improved developmental and participation outcomes associated with participation in rehabilitation services, it is critical that families who chose to engage in children's rehabilitation have adequate access to services available to them. Of notable concern, is when a family experiences barriers to accessing rehabilitation services as a result of factors such as the age of a child, parental stress, and culture or socio-economic status (SES) (19, 20). Participation in therapy may be limited when personal and environmental factors create barriers to service use. For example, language barriers and navigating unfamiliar health systems have been identified by immigrant mothers as barriers to accessing available services for their child with a disability (21), which would have negative implications for participation in children's rehabilitation. At a health systems level, families who experience barriers (e.g., transportation, working hours of parent) to consistently attend appointments may be systematically excluded from participating in rehabilitation by policies, which result in families being discharged after missing a specified number of visits (22). The personal and environmental factors should be considered with respect to the societal influences and systemic inequities that may limit therapy participation for some children and families (23). Systemic inequities are defined as disparities in health outcomes as a result of the uneven distribution of power, goods, and services (24, 25). This perspective study describes how an intersectional lens can be applied to critically examine the potential impact of systemic inequities on participation in therapy for children with disabilities.

Grounded in a biopsychosocial model of disability, the ICF acknowledges the impact of environmental and personal contextual factors on the experiences of the participation of individuals (1, 2, 26, 27). In the ICF, the term environment is used broadly to represent the physical, social, attitudinal, and institutional context in which a person is situated (1, 2, 12, 28). The environment has been demonstrated to influence experiences of participation, with the potential to act both as a facilitator or barrier to participation for children with disabilities (6, 7, 28, 29). Parents of children with disabilities described features of the environment as making it harder for their children to participate in community-based activities (7), including participation in rehabilitation services. The potential influence of the social environment on participation was demonstrated in a systematic review examining the impact of family factors such as family structure, socio-demographic factors, parental behavior, and family resources on participation outcomes for children with disabilities, both generally and with specific reference to accessing the rehabilitation services of children (19, 29).

The ICF framework identifies personal factors that have been shown to affect participation such as gender, age, and ethnicity (1, 2). These personal factors contribute to the make-up of the unique identity of a person and are distinct from the disability or health condition (1). Individual categories of personal factors are not defined in the ICF (1). Examining the need for more specificity within the ICF personal factors has been identified in the literature (30, 31); however there is concern that classifying personal factors with single categorical distinctions risks discrimination and misrepresenting the personal factors with which a person does or does not identify (32–34). When single-identity categories (e.g., age, gender, or ethnicity) are considered in isolation service providers may lack sufficient information to set tailored goals collaboratively with clients, risk-making assumptions about how clients position themselves in relation to their personal factors and underestimate the environmental opportunities and challenges that may impact participation resulting from the person-environment relationship. Therefore, this paper proposes intersectionality theory as a means of bridging the understanding of the ICF personal and environmental factors that can impact participation in therapy. By applying an intersectional lens, all facets of the identity of a child are considered simultaneously, with specific acknowledgment for the environmental context in which a child with a disability is situated.

Intersectionality theory can be used to holistically identify and critically examine the aspects of identity by exploring the relationships that exist between facets of the identity of an individual (i.e., ICF personal factors) and the larger societal systems in which a person participates (35). Intersectionality allows for the application of a critical lens to examine how the ICF personal factors as experienced by a child with a disability contribute to whether society views them as belonging to groups of socially-perceived advantage or disadvantage. This social construction of the identity and place of a child in the society has implications for their participation in therapy. It is important to note that in this context the term critical is used to describe the process of thinking deeply about the intended and unintended consequences associated with our actions (36). Intersectionality theory explains that identity cannot be understood by examining individual elements of identity (37). Instead, we examine the socially constructed privilege and oppression associated with the interaction between multiple aspects of the identity of an individual and their environments (37, 38). Literature and frameworks related to childhood disability and participation illustrate the relationship between factors external to the child (ICF environment factors), factors internal to the child (ICF personal factors), and participation outcomes (12, 26, 28). The family of participation-related constructs model embeds participation within the surrounding environmental contexts and makes explicit the bidirectional relationship between factors intrinsic to an individual and participation (28). In childhood disability literature, next steps should include a critical exploration of how the personal factors of an individual are privileged or oppressed, impacting their life participation. For example, it is understood in the literature that the environmental context mediates participation frequency and level of involvement in activities (6). Given that many personal factors cannot be changed, modifying the environment has been discussed as an approach to facilitate participation (6). Critically examining the social, attitudinal, and institutional environments in which children with disabilities are situated creates opportunities to identify barriers to life participation. As a result, participation-enhancing solutions, focused on modifying the systemic environmental context in which inequities exist, can be developed. Intersectionality theory provides the critical lens needed to examine contextual factors identified using the ICF to examine potential systemic inequities impacting participation.

CRITICALLY EXAMINING PARTICIPATION IN CHILDHOOD DISABILITY

Children with disabilities have unique identities, in part, shaped by the physical, social, attitudinal, and institutional environments around them (39). The social environment referenced in the ICF includes the family of a child. Parents are the most proximal environment to a child, playing a critical role in facilitating opportunities for participation and providing care for children with disabilities. However, the broader environments in which the family and child are situated need to be considered (40) to understand the implications for participation in children's rehabilitation. Personal factors such as age, sex, or ethnicity do not alone determine therapy participation. Instead, implications for the participation of a child arise when the interplay between their unique personal factors and the broader environmental context results in experiences of systemic inequities such as ageism, sexism, racism, or ableism. Experiences of discrimination risk limiting a child with a disability from fully participating in important aspects of their lives, including rehabilitation therapies. Applying a critical intersectional lens to participation in therapy allows clinicians to holistically consider how the identity of a child interacts with the surrounding environment to better understand implications for participation. Opportunities for participation in therapy are created or repressed according to the complex interaction between a the personal identity factors and the systemic inequities of a child that exist in rehabilitation environments.

Examining the personal factors of a child individually does not provide an adequate foundation for understanding implications for participation in pediatric rehabilitation services. Applying an intersectional lens allows us to critically consider how personal factors, as outlined by the ICF, interact with the ICF environmental domains to maintain systemic inequities and impact participation in therapy. This creates an opportunity for the rich examination of potential facilitators and barriers to participation. As an example, let us briefly explore the personal factors of sex and culture in relation to disability. In some developing countries there continues to be a gap in participation opportunities between boys and girls in life activities such as schooling and employment (41–43). Additionally in some cultures, stigmatization of disability prevents parents from seeking therapy services for a child with a disability (44, 45). In this context, opportunities for participation, including participation in therapy, for a girl with a disability may be limited due to the possibility of sexist and ableist discrimination resulting from person–environment interactions. This example demonstrates the need to explore the intersection between the personal factors of sex and culture as influenced by disability and the environment in which the family is situated to understand implications for participation in therapy. This example illustrates how intersectionality theory can be applied in complement to the ICF by contributing a critical lens to examine the interaction between the ICF personal (i.e., sex, culture) and the environmental factors (i.e., stigma) to identify potential participation restrictions resulting from systemic discrimination. Although this study focuses on implications for participation in therapy, this perspective can be applied when examining how children with disabilities participate in a variety of life contexts.

DISCUSSION

Applying a critical perspective facilitates an in depth understanding of how the person–environment relationship potentially impacts the participation of children with disabilities in rehabilitation services. But what do we as childhood disability clinicians do with the insights gleaned from critically examining the interplay between the intersectional identity and surrounding environment of a child? How can this information be used to optimize participation and inclusion of this population in childhood rehabilitation? By considering how the personal and environmental context of a child might impact their participation in therapy, clinicians have the opportunity to conceptualize solutions to enhance access for an individual family as well as identify patterns in participation limitations that could drive system-level change. At the clinical practice level, the use of a family-centered and solution-focused approach, whereby the family is actively engaged as collaborators in therapy, may be a way for clinicians to gain an understanding of how the person–environment relationship creates barriers to therapy participation and involve families in developing solutions (46, 47). A family-centered approach to care recognizes the expert knowledge of a parent about their child and has been associated with improved access and health outcomes in children with special needs (47, 48). Therefore, a family-centered approach to identifying and co-creating individualized solutions addressing parent-identified barriers to participation in children's rehabilitation is recommended. Inclusion of diverse stakeholders has been identified as critical in policy development (49). Clinicians can advocate for the representation of the family voice in the development of policy that supports inclusive participation in therapy.

As a critical approach, intersectionality seeks not only to understand lived experience of others and highlight oppression, it also aims to generate new knowledge that calls for change to inequitable social practices (37, 50–52). Clinicians are well positioned to become allies, working alongside families and children to understand their experiences of being included and excluded from opportunities to participate in therapy and identify potential inequities resulting from the person–environment relationship. This information provides a platform to highlight the role of society in facilitating or hindering participation in rehabilitation services and advocate for system-level changes, such as resource allocation, program, and service design or policy reform that optimize inclusion. The discomfort clinicians may experience while acting as critical allies is important to acknowledge. On one hand, equity is a core component of health ethics and should be advocated for (14, 53) but on the other hand, clinicians have a commitment to follow the rules and practices of the health system by which they are employed. Critical allyship may require clinicians to advocate in opposition to the dominant policies and practices of the system they work in. Although there is no clear solution to address the paradox created by critical allyship, clinicians can make use of frameworks such as the 7-step framework for critical analysis to reflect on the impact of their actions in practice and consider the potential harms and benefits associated with different courses of action (36).

There is a need for careful consideration on how clinicians align themselves and act upon this advocacy role to avoid employing a disempowering approach aiming to "fix" those in a position of socially constructed disadvantage (i.e., children with disabilities) (54). In the coin model of privilege and critical allyship, Nixon (2019) describes "practicing critical allyship" as an approach for individuals in a position of privilege (i.e., clinicians) to work in partnership with those experiencing oppression to identify and take action on the systems perpetuating inequities (i.e., restrictions in therapy participation). Critical allyship calls individuals in positions of power to acknowledge their experiences of privilege and how their advantaged position may contribute to sustaining dominant inequitable social practices (54). As critical allies, clinicians can learn from parents and children about their experiences to better understand the impact of system inequities on participation in children's rehabilitation (54). Additionally, under the guidance of parents and children, clinicians can use their privileged positioning to advocate for system change among other power-privileged groups (i.e., health service decision makers) (54).

By applying an intersectional lens to examine the impact of the person–environment relationship on participation in therapy for children with disabilities, clinicians have the opportunity to practice critical allyship alongside children and parents to transform inequitable systems. However, applying a critical lens to participation in therapy may be a new approach for some clinicians. How as a group of professionals do we implement and become comfortable critically examining participation outcomes for children with disabilities? We suggest looking toward the ICF as a framework to get started. The ICF is a well-recognized, familiar, and frequently referenced framework in the childhood disability literature (26, 55–57). Through listening to the thoughts and feelings shared by families of children with disabilities, clinicians can apply the ICF to conceptualize the personal and environmental factors relevant to the individualized family context. Literature is available to provide clinicians with pragmatic guidance for integrating the ICF into their practice (58). Critical examination of how the dominant ways of thinking in our society impact participation in therapy is currently under-represented in the literature. An intersectional lens can be used in complement the ICF to critically examine contextual factors, identify barriers and facilitators to participation in children's rehabilitation, and create actionable change toward more inclusive systems.

DATA AVAILABILITY STATEMENT

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

AUTHOR CONTRIBUTIONS

MR, LL, BD, and MP contributed to the conception of this work, contributed comments and edits to the initial and subsequent versions of the manuscript, read, and approved the final manuscript. MR wrote the first draft of the manuscript. MP is the senior author of this manuscript. All authors contributed to the article and approved the submitted version.

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REVIEW
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Canadian Resources for Siblings of Youth With Chronic Health Conditions to Inform and Support With Healthcare Management: A Qualitative Document Analysis

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Background: As children and adolescents with a chronic health condition (CHC) age and transition to adulthood, many will increasingly assume responsibilities for the management of their healthcare. For individuals with CHCs, family members including siblings often provide significant and varied supports. There are a range of resources in Canada to support siblings of individuals with a CHC, but these resources are not synthesized and the extent to which they relate to healthcare management remains unclear.

Purpose: The purpose of this document review was to identify, describe, and synthesize the types of resources currently available to provide general information and healthcare management information about how siblings can provide support to individuals with CHCs in Canada.

Methods: Print and electronic resources were systematically identified and retrieved from the websites of organizations, treatment centers, and children's hospitals that are part of Children's Healthcare Canada. Each unique resource was treated as a text document. Documents that met the following inclusion criteria were included: addressed the topic of siblings of individuals with a CHC and written in English. Data were extracted from included documents and qualitative conventional content analysis was conducted. Throughout the process of this review, we partnered with a Sibling Youth Advisory Council.

Results: The systematic search yielded 1,628 non-duplicate documents, of which 163 documents met the inclusion criteria. Of the total of 163 documents, they were delivered in the following formats: 17 (10%) general informational products (e.g., booklets, videos) about a CHC and sibling relationships, 39 about support programs and workshops (24%), 34 news articles (21%) that described the roles of siblings, and 6 (3%)

healthcare management informational products (e.g., toolkit, tipsheets), 31 blogs (19%) and 39 interviews (24%) with parents and siblings. In the blogs and interviews, siblings and parents described how siblings developed knowledge and skills for healthcare management, as well as their role and identity over time.

Significance: This study identified that there are limited resources available about healthcare management for siblings of CHC in Canada. Resources are needed to facilitate conversations in the family about the role of siblings with healthcare management of their sibling with a CHC.

Keywords: chronic health condition, siblings, qualitative analysis, pediatrics, family-centered services, healthcare management, transition to adulthood

INTRODUCTION

In North America, ∼15 to 18% of all youths have a chronic health condition (CHC) (1, 2). The term "chronic health condition" encompasses congenital and acquired diseases, as well as physical and mental health conditions (3). There has been a shift toward providing care for a family of a child with a CHC using a non-diagnostic approach (4). Instead of focusing on a specific diagnosis or condition alone, increasing care, and supports focus on providing comprehensive care to address the holistic needs of the individual and family (4, 5).

Families often express significant concerns about how they can best support their child during the transition from the pediatric to adult healthcare systems (6). During this time, youth with CHCs will need to learn how to manage their healthcare, for example learning how to navigate the process of filling prescriptions, scheduling healthcare appointments, and answering questions from healthcare providers. Typically, support is provided by family members through this transition period. In addition to healthcare management, individuals with CHCs are also exploring their interests and goals, including school, work, and leisure (6–9) and learning to navigate new environments, including healthcare for adults, education, transportation, recreation, and social services (7). Throughout the lifespan, families typically can provide support, given their past experiences coordinating their family member's or child's care and knowledge of their child's strengths, areas of improvement, and goals (10).

In addition to parents, siblings are a part of the family who can provide support for their sibling with a CHC. Within the typical lifelong bonds between siblings, these relationships are highly dynamic and can change over time depending on the needs, roles, and commitments of the whole family (11–13). Each sibling relationship is different with varying levels of emotional closeness, social connectedness, and expectations of each other (14). During childhood and adolescence, sibling relationships are unique as they often live and grow up in their shared home environment where they can act as peers, confidants, or role models (15). At a young age, siblings of individuals with a CHC often recognize that they need to support their family in different contexts (16, 17). When there is future planning involved from the whole family, siblings often feel closer to each other and with their family, and they have a clearer understanding about their role for the future during adulthood (18, 19).

In some families, there may not be discussions about the role of siblings but siblings may be expected to become carers to their sibling with a CHC (20). In 2018, the Siblings Needs Assessment Survey was conducted in Canada among young adults who were ages 20 years or older, who had a sibling with a disability and received a total of 360 responses (21, 22). Siblings described concerns for their sibling's future such as finding employment or living independently (12, 23). Siblings might have worries for new responsibilities, such as guardianship or financial responsibilities, when their parents can no longer be the primary caregivers (13, 24, 25). These concerns can affect the extent to which siblings are involved in the healthcare of their sibling with a CHC.

Typically developing siblings might want to support their sibling with a disability, but they require knowledge and skills on how to do this. There are currently "Sibshops" that are offered across ten countries, including the United States and Canada (26). These Sibshops provide an opportunity for siblings to connect with people with similar experiences and share stories. A survey was conducted to evaluate Sibshops, in which 66% of respondents identified that they learned coping strategies, 75% reported that Sibshops had a positive impact on their adult lives, and 94% stated that they would recommend Sibshops to others (27). Often, one of the goals of Sibshops is to provide a space for siblings to meet and share experiences with other siblings of individuals with a CHC in a recreational setting (28). Some SibTeen sessions are also held for adolescents ages 13–17 years old to offer a community of support (29). Although there are support groups for siblings of individuals with a CHC, such as Sibshops and SibTeen sessions, there are no tailored resources or programs for typically developing siblings to share their concerns about supporting their sibling with a CHC specifically for healthcare management. There are many ways that siblings can provide support to their sibling with a CHC. These can be categorized as: concrete support such as taking on responsibilities and providing assistance; emotional support such as listening and empathizing; advice support such as offering information; and esteem support, such as expressing encouragement (30). Siblings can offer these different types of supports to help their sibling with a CHC manage their healthcare.

Informational needs have been identified to be a critical need for siblings (31). Siblings who wish to have a caregiving role often seek knowledge in how to provide care to their sibling with a CHC, how to navigate disability services, and how to seek supports for themselves (31). While there is information available and advertisements of services for siblings on websites of children's hospitals and organizations, many families and siblings identified that they were not aware of this information (32, 33). Siblings identified that they want to have an open, constructive dialogue with their parents about the future, including expectations and responsibilities (13, 24). Often, siblings had to learn how to care for their sibling on their own as information was not always passed down from parents to the siblings (11, 24).

Informational needs are also increasingly being addressed by individuals, including siblings and their families, through the use of the Internet. Siblings can share their experiences and needs online in various formats, such as blogs. Among the few studies that have analyzed the content of blogs, researchers identified how individuals who write these blogs can share experiences that might be different from what might be shared in a research study. Young adults and families have previously written blogs to document their experiences in healthcare, including their emotions and challenges (34–36). Similarly, blogs written by siblings and families can provide insights into the needs of siblings in order to prepare for their roles with healthcare management. There is a gap with little information known about the types of needs about healthcare management that siblings of individuals with CHC are sharing online.

Individuals may also choose to find information online for various reasons, including medical information, such as options for therapy, treatments and health services (37, 38). Siblings of individuals with a CHC require information on how they can provide support with healthcare management. In the Canadian Sibling Needs Assessment Survey, the majority of respondents across all age groups identified online websites as their preferred method for resources, information and tools (22). Programs, such as Sibshops and SibTeen sessions in North America, are often promoted online and share information about eligibility criteria and registration. In other countries, initiatives to support the needs of siblings of individuals with a CHC include Siblings Australia developed in 1999 (39), Sibs in the United Kingdom in 2001 (40), and the Sibling Leadership Network in the United States in 2007 (41). These initiatives in Australia, the United Kingdom, and the United States have been established for many years, and includes an array of support programs and resources for siblings of individuals with a CHC. In Canada, the Sibling Collaborative was established in 2017 and offers online support groups with some resources such as information about the COVID-19 vaccine, finances, and stories from siblings (42). Despite the availability of many resources, as a team, we have heard from siblings that resources about healthcare are not easily accessible or retrievable in Canada. The resources are often posted on certain websites by children's hospitals and organizations, but the websites are not easy to navigate. In Canada, there is no national systems approach to store resources for siblings of individuals with a CHC. Considering the important and multi-faceted roles that siblings can have, it is important to identify and summarize the different types of resources that are available to siblings of individuals with a CHC.

This review aims to identify and describe:

i. the types of resources currently available in Canada to provide both general information and specific healthcare management information about how siblings can provide support to their sibling with a CHC; and
ii. key topics discussed in resources created by siblings and families.

METHODS

Integrated Knowledge Translation

An integrated knowledge translation approach was used throughout the process of this review to partner with the Sibling Youth Advisory Council (SibYAC) comprised of six young adults who have a sibling with a disability. The SibYAC were first involved with the idea and concept, as well as the research question of this review. The SibYAC shared their experiences with searching for information to support their roles as siblings, and they identified a need to identify and synthesize resources that are available to siblings of individuals with a CHC. These experiences from the SibYAC provided a clear rationale to support our review aims. There were individual check-in meetings with each SibYAC member, and an engagement framework (43) and Involvement Matrix (44) were used as tools to ask about the tasks and roles that they would like to have in this review. The SibYAC were further involved in data analysis by sharing their perspectives for the retrieved documents to ensure that the extracted data are synthesized meaningfully for siblings, families, and other stakeholders. They were then involved with the interpretation of results and drawing conclusions. Meetings were held with the SibYAC to ask about their reflections of the summary of results with guiding questions including: How do the documents and websites support siblings in their role? Based on the documents and websites, what are some needs, information, or questions that you still have as a sibling? For example, in healthcare or in general. Reflections from the SibYAC helped to identify the gaps and future directions about resources to support siblings in their roles, including with healthcare management of their sibling with a CHC.

Qualitative Document Analysis

Qualitative document analysis involves a systematic search of documents and resources, which includes both printed and electronic resources (45). A variety of documents can be analyzed, including books, brochures, diaries, journals, event programs, or news articles.

Search Strategy

A comprehensive search was conducted on publicly available websites of thirty-one organizations, including children's hospitals, and rehabilitation centers that are part of Children's Healthcare Canada (46). These were selected to provide an initial understanding about the types of resources that are available for siblings of individuals with a CHC in a healthcare setting.

The websites were searched in August 2020. A broad search strategy was employed in the search engine of each website with the terms: "sibling," "brother," or "sister." All documents from the search were digitally retrieved using a feature called NVivo Capture and imported into NVivo (Version 11.4.3). Duplicates of documents across websites were removed.

Inclusion and Exclusion Criteria

Text from all retrieved documents was initially scanned in NVivo for the key terms of "sibling," "brother," or "sister." Documents that included at least one of these key terms were read by the first author (LN). Identified documents were included in the review if they: (1) addressed the topic of relationships between siblings with and without a CHC; and (2) were published in English. Documents were excluded if the sibling was mentioned but did not discuss supports of siblings or the relationship between siblings of individuals with a CHC.

Ethical Considerations

Ethics approval was not required to retrieve and analyze documents that are publicly available on the Internet. An assessment of online documents can be conducted to identify the intent of online documents and its use in research, and documents that are written for public intent do not require consent from the creators or authors of the documents (47). In the analysis of retrieved documents, there was careful consideration to protect the privacy of the creators for the documents, and all personal identifiers were removed from included documents.

Data Extraction and Analysis

A data extraction template was created using Microsoft Excel Version 16.41 to collect data from each document (48). This template included the following categories: document source, document type, purpose/goals, and key content. For document types coded as "blog" or "interview," content data for two additional categories were extracted: (1) family characteristics; and (2) CHC of an individual in the family. Additionally, all blogs and interviews were read and re-read in an iterative process to achieve immersion in the data and understand the stories shared by siblings and parents. Conventional content analysis was conducted by the first author (LN) for documents that were coded as blogs or interviews (49). Initial codes were developed based on the full text of the blogs and interviews, and these codes were then organized into categories to depict how they were related and linked to each other. Codes were grouped into meaningful clusters or categories based on their similarities in concepts. An Excel spreadsheet was created, that included extracted quotes and codes that were grouped into categories. Each category was expanded into a short statement to describe the key topic shared by siblings and families. Two analysis meetings were then held with individuals familiar with the content (e.g., SibYAC) and qualitative analysis (e.g., graduate students, co-author SJ) to review and name the categories, and identify additional properties and dimensions of each meaningful cluster. Analytic notes were written by the first author (LN) about how the categories related to each other to form meaningful clusters. While the content of all documents was analyzed to identify information and supports for healthcare management of an individual with a CHC, conventional content analysis allows for the identification of key topics from included documents that describe the experiences of siblings of individuals with a CHC beyond healthcare management. In this review, recognizing that gender is non-binary, we refer to siblings as a "brother" or "sister" based on the information provided in the resources included in this review with the recognition that siblings may identify themselves along a spectrum.

Data Credibility

To ensure credibility of the data, an audit trail and multiple analyst triangulation were used as two strategies. An audit trial was created to describe the steps and document decisions that were made about data extraction, as well as the identification of codes, categories, meaningful clusters, and key topics identified in the documents (50, 51). Sufficient time was also spent reviewing each source of information to identify recurrent patterns and key topics of the documents (52). The first author (LN) spent extensive time to read and re-read all documents, and took field notes of emerging ideas for each document in an Excel document (e.g., What is the main message about this document? How does this document relate to other documents?). To further enhance the credibility and dependability of the data, the lead author engaged in reflexivity and documented their own biases, preferences, and preconceptions about the topic in a series of memos (53, 54). Analyst triangulation was employed, in which multiple individuals with different backgrounds and expertise offered their perspectives about the preliminary and final findings (54). Two initial meetings were held to review and discuss how to organize preliminary findings: first with a group of graduate students with expertise in mixed methods and qualitative research, and then with the SibYAC. Two additional meetings were held with the SibYAC to share their reflections about the meaning of the findings in this review to them, describe whether the key topics from the blogs and interviews resonated with, or differed from, their experiences as young adult siblings of individuals with a disability, and identify gaps for future directions. All SibYAC members present at the meeting described that the key topics were similar to their experiences, and they provided suggestions for future directions in the development and enhancement of resources for siblings of individuals with a CHC. All authors of this review are from a multidisciplinary backgrounds including cognitive psychology, education, nursing, occupational therapy, physiatry, rehabilitation, patient-oriented research, and lived experiences, and all provided their perspectives on the synthesis of findings.

RESULTS

The systematic search yielded 1,628 non-duplicate documents and resources, with 1,015 documents and resources that included keywords of "sibling," "brother," or "sister." There were 163

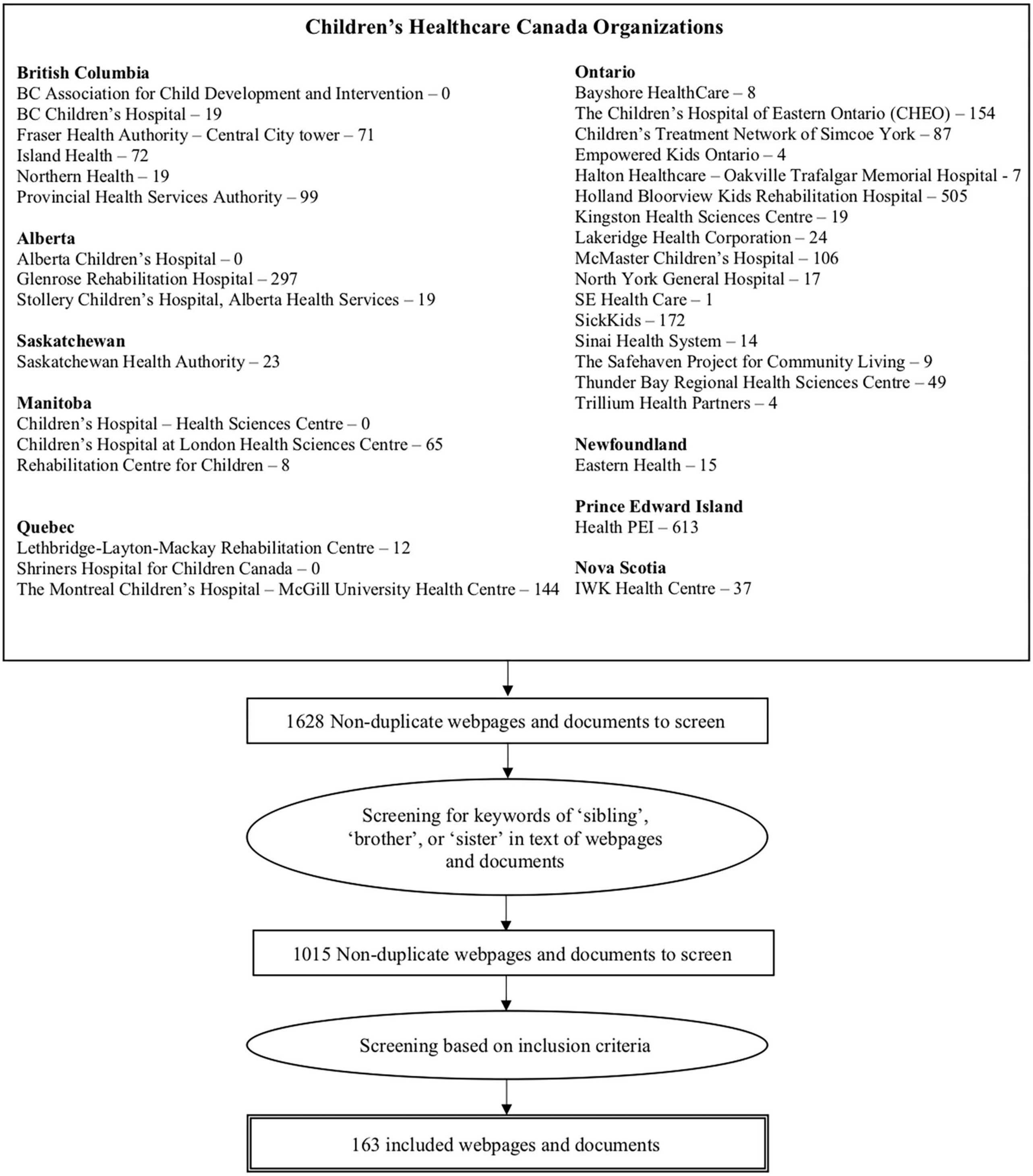

FIGURE 1 | Flow diagram outlining the section of included websites and documents.

documents and resources that met the inclusion criteria (See **Figure 1**).

All resources were identified from treatment centers that provide inpatient and outpatient services to children and adolescents with a CHC. Some documents discussed CHCs as a broad group of conditions, while others referred to specific conditions such as autism spectrum disorder, cerebral palsy, Down syndrome, epilepsy, genetic disorders, juvenile arthritis, intellectual disorders, or mental health disorders. The documents included 6 books (55–60), 1 podcast (61), 34 programs and 5 workshops [references provided to websites where ongoing programs (62–67) and workshops (63) are advertised], 7 films and videos (68–74), and 34 news articles. Among the 34 news articles, they referred to 2 announcements (75, 76), 6 "awareness" recognition of different days or months of CHCs (77–82), 5 events (82–86), 10 research studies (87–94), 4 participation in research [references provided to websites where active research studies are posted (95, 96)] and 7 stories (97–103). Three documents referenced a toolkit (104–106), 3 booklets (107–109), and 3 tip sheets (110–112). Of the total of 163 documents, there were 31 blogs (19%), of which 13 (8%) written by parents and 18 (11%) written by siblings, and thirty-eight interviews (23%) with 12 (7%) interviews with parents and 26 (16%) interviews with siblings, and one interview with a family that included both the parents and siblings. **Table 1** provides details about the document type, source of documents, number of documents and references, target audience, purpose and goals, and summary of key content.

Types of Resources for General Information

The majority of resources were general informational products for siblings of individuals with a CHC, which are available in a variety of formats. **Supplementary Table 1** presents detailed descriptions of these resources.

i. *Booklets and books.* Books were available for siblings and families, in which some highlighted the need to understand the importance of sibling relationships, for example, creating a space for siblings to understand their emotions when they have a sibling with a CHC. Booklets were also available to provide guidance to parents and teachers about how to communicate with siblings of someone with a CHC.
ii. *Podcasts.* Personal stories from families, including siblings, were shared through podcasts, films, and videos. These stories described the journey of the whole family, and one podcast discussed the relationship between the siblings in which one sibling has a CHC.
iii. *Programs and workshops.* There are advertisements that announced past programs ($n = 34$) and workshops ($n = 5$) available to siblings and families. Among these 39 documents, 17 were in-person, 8 were virtual due to COVID-19, and 14 did not indicate the type of format. Most programs offered were sibling support groups, such as Sibshops, that are available throughout the year for siblings who are ages 7–25 years old. There were programs specifically for families to connect with other families of children with autism spectrum disorder available for free [reference to ongoing advertisement about the program (57)].
iv. *News articles.* All stories that were published as a news article were authored either by parents ($n = 1$), mothers ($n = 2$), or a sibling ($n = 3$), both a mother and sibling ($n = 1$). Articles authored by mothers focused on stories of their child's lived experience, parenting multiple children with a CHC or the same CHC, and/or the roles that other children may assume when there is a child with a CHC. Siblings discussed topics, such as sharing their emotions about their sibling relationship, providing support with healthcare management, and transitioning into different roles as a sibling, such as becoming a caregiver. Throughout the year, there were news articles with announcements about initiatives that were inspired by the stories of siblings. For example, there were announcements about various "awareness" days and months about specific disabilities and health conditions, which provided an opportunity for siblings to share stories about their sibling with a CHC (69–74). News articles also advertised research studies that were completed or actively recruiting sibling participants. The topics of these studies included genetic studies for specific health conditions, such as autism spectrum disorder and lymphoma, successful organ transplants between siblings, effectiveness of assistive equipment, and a survey to understand the needs and feelings of siblings of youth with a CHC.

Type of Resources for How Siblings Can Provide Support With Healthcare Management

There are few resources that provided information for siblings about their roles with respect to the healthcare management of their sibling with a CHC. When resources were available, they were formatted as either tip sheets or as a toolkit.

Toolkit and Tip Sheets

Both parents and siblings could refer to different sheets that were available for download online, which included tip sheets (110–112), and a toolkit (104). These sheets also focused on strategies for how siblings can provide support to their sibling with a CHC. For example, there was a tip sheet that described strategies for siblings of inpatients at a children's hospital (112). Some strategies for how siblings can be included as part of the inpatient stay were being a part of their sibling's care team, doing fun activities together, talking to their sibling, and helping the sibling to decorate their room (112). While the resources primarily focused on providing knowledge about a CHC to siblings, some resources provided additional strategies for siblings to support the healthcare management of their sibling with a CHC. A toolkit was also co-designed with siblings, clients, parents and clinical staff for brothers and sisters of children who have an acquired brain injury (104). The toolkit was described as a resource that siblings can use to learn knowledge about their sibling with an acquired brain injury and learn how to explain this injury to other adults who can provide help, when needed (104).

Blogs and Interviews

Siblings and families described different types of CHCs in blogs and interviews. Some siblings also had the same CHC as other siblings in the family. The age of siblings and individual with a CHC discussed in blogs and interviews ranged from infancy to older adults. The size of families ranged from one to five children. Based on an analysis of the content from blogs and interviews shared by siblings and parents, a conceptual map was developed

TABLE 1 | Description of documents.

Document type	Source of documents	Number of documents and references	Target audience for resource	Purpose and goals	Summary of key content
Resources about general information					
Booklets	One hospital in Ontario and one service provider in Prince Edward Island	3 (107–109)	Parents and educators of an individual with a CHC	To offer strategies for parents and educators of an individual with a CHC and their siblings	Two booklets, one for parents and one for educators of individuals with autism spectrum disorder, included a booklist relevant for siblings to learn about autism. One booklet offered strategies for educators to support siblings of individuals with childhood cancer
Books	Two hospitals in Ontario	6 (55–60)	Public	To share about the importance of relationships of siblings, when a sibling has a CHC	The books recognize how there is a needed space and role of siblings
Podcast	One hospital in Ontario	1 (61)	Public	To tell the stories of people with a CHC, including the experiences of siblings in families with a child with a CHC	One podcast spoke about the stories of siblings of people with a CHC
Programs	Four hospitals and two service providers in Ontario	34* (62–67)	Parents/caregivers and siblings of an individual with a CHC	The purposes of the programs included providing opportunities for families, including siblings, to connect with other families of individuals with a CHC, spend together as a family through various community events, providing support to siblings to understand the CHC and develop coping strategies	Programs included: • Family Nights held on a regular basis (e.g., monthly) for siblings and families of individuals with a CHC to connect with each other • Informal family playgroups that have regular events (e.g., monthly) for families to participate in the community, e.g., indoor playground, escape room, arts and crafts, board games • Support program, including the Sibshops with sessions for siblings with a range of ages from 7–25 year olds to support siblings who may have questions or are seeking coping strategies with their brother or sister with a CHC • Bravery Beads Program that allows children an opportunity to collect a different bead for each procedure or event while visiting the hospital for a treatment. An example of how the beads were used between siblings, where the sister educated the class on what her brother went through and still has to go through
Workshops (one-time event)	One hospital in Ontario	5* (13)	Siblings and parents of a child with a CHC	The purposes of the workshops were to provide opportunities for siblings to connect with other siblings, discuss ways that parents can support siblings of individuals with a CHC in the family, and advertise other ongoing programs for siblings and families	An opportunity for parents and siblings (either in separate groups or together) to ask questions, including to a panel of adult siblings about their experiences. Workshops were provided both online and virtually

(Continued)

TABLE 1 | Continued

Document type	Source of documents	Number of documents and references	Target audience for resource	Purpose and goals	Summary of key content
Films and videos	One hospital in Ontario	7 (68–74)	Public	To share about the experiences of families, including the perspectives of siblings and parents, about growing up with a child with a CHC	The types of CHCs covered in the films and videos included autism spectrum disorder, Down syndrome, brain damage, and Type 1 diabetes
News articles					
Announcements	One hospital in Ontario	2 (75, 76)	Public	To share information about results, which included a donation to create a center in Canada to support adults with disabilities, a partnership to build rehabilitation capacity for children with a CHC between institutions, and the winner of a "filmpossible" award	Donation and partnership were inspired by the experience of a family with an individual with a CHC. Stories about the experiences of siblings of an individual with a CHC shared in the announcement
Awareness about CHCs	Two hospitals in Ontario and one hospital in Quebec	6 (77–82)	Public	To raise awareness on specific days to appreciate different disabilities and roles, in which siblings have a part in the awareness of CHCs	Each document described the awareness of different days: • Sibling Appreciation Day • Purple Day for Epilepsy • Cerebral Palsy Awareness Month • Childhood Cancer Awareness Month • Children's Grief Awareness Day • International Day of Persons with Disabilities To raise awareness of these different days, siblings shared stories about their brother or sister with a CHC
Events	One hospital in Ontario and one hospital in Quebec	5 (82–86)	Public	To raise awareness about the stories and roles of siblings	Events included advocacy for individuals with disabilities, as well as events for siblings to attend
Research Studies	Two hospitals in Ontario and one hospital in Quebec	10** (87–94)	Public	To share the findings of research studies	The topics of research studies included genetic information for siblings of individuals with ASD, genetic mutation that lead to lymphoma, survey findings about the experiences of siblings of a brother or sister with a developmental disability, successful donor liver transplant between siblings, rare autoinflammatory disease based on research of two siblings with juvenile idiopathic arthritis, immunotherapy, trials about accessible equipment, and accessible video games for children with disabilities
Participation in research	One hospital in Ontario and one health center in Nova Scotia	4* (95, 96)	Public	To recruit participants for a research study	The topics of the research studies were assessments of early behavioral signs of autism spectrum disorder in infants and the experiences of sibling including their needs and feelings

(Continued)

TABLE 1 | Continued

Document type	Source of documents	Number of documents and references	Target audience for resource	Purpose and goals	Summary of key content
Stories	Health service provider in British Columbia, three hospitals in Ontario	7 (97–103)	Public	To share the stories of families who have an individual with a CHC	Stories were shared by mothers and siblings about their experiences with a person with a CHC. For some sibling relationships, both siblings had a CHC, and shared how they supported each other
Resources for how siblings can provide support with healthcare management					
Toolkit	One hospital in Ontario	3 (104–106)	Siblings of an individual with a CHC	To offer strategies for siblings of individuals with an acquired brain injury	All documents referred to one toolkit, the SibKit 1.0 (105), which provides information about strategies for siblings of individuals with an acquired brain injury
Tip sheets	One hospital in Ontario and one hospital in British Columbia	3 (110–112)	Parents and siblings of an individual with a CHC	To offer strategies for parents to support siblings, and for siblings to support their sibling with a CHC	One tip sheet described how parents can support siblings of an individual with a CHC. Two tip sheets offered strategies for siblings to cope with the surgery of their brother or sister, as well as, when their brother or sister is an inpatient
Interviews					
With Siblings	Three hospitals in Ontario and one hospital in Quebec	26***	Public	To share about the experiences of siblings of a child with a C	Key topics discussed during the interviews and blogs are presented in **Table 2**
With Parents	One hospital in Ontario	12***	Public	To share about the parents' experiences when they have multiple children including a child with a CHC	
Blogs					
Written by siblings	Three hospitals in Ontario	18***	Public	To share key messages from siblings about their experiences when they have a sibling with a CHC	
Written by parents	Two hospitals in Ontario and a health service provider in British Columbia	13***	Public	To share the stories of families of a child with a CHC and the siblings from the parents' perspective	

**Programs, workshops, and participation in research studies are advertised and updated on an ongoing basis on the websites of the children's hospital and/or treatment centers. The number of documents refers to the advertisements and newsletters that was posted on the websites.*

***Two documents are no longer available on the website, but was included in the analysis.*

****References were not provided in order to ensure confidentiality of the authors and families mentioned in the blogs and interviews.*

to describe the codes, categories, and key topics discussed in these documents (See **Figure 2**). Detailed descriptions about these key topics are described in detail below. The frequency that these topics were identified in the blogs and interviews are provided in **Table 2**.

Siblings' Development of Knowledge and Skills for Healthcare Management

Siblings described how they needed to learn about the meaning of disability. Some siblings did not understand specific CHCs, such as the different treatments and services their siblings had to receive to manage their CHC. Siblings often described that they simply saw their sibling for who they were, regardless of the CHC. Parents shared stories in their blogs about the forming of relationships between their children. Young children learned how to develop their relationship with their sibling with a CHC. A mother shared the story of how she saw her two children interact with each other, where her young daughter asked to hug her brother or hold his hand when he was using an assistive device to walk. As siblings began to develop an understanding about the CHC, some siblings offered support with healthcare management. For example, a mother described how her daughter learned to be present and hold her brother's hand when he was using a suction machine. Siblings shared in interviews about how they learned different ways to support their siblings. For example, a sister observed her mother apply breathing techniques with her brother and she learned how to do the same. There was a process in which siblings first needed to learn about the CHC and develop a relationship with their sibling with a CHC, which then allowed them to learn how to offer support with healthcare management.

Siblings' Development of Role and Identity

The role of being a sibling to someone with a CHC provided them with experiences about a CHC, and the sibling role became a part of their identity. Young adult siblings shared in written blogs about how they were developing their own identity, such as moving away for university and developing their career. For some siblings, the experience of growing up in a family of an individual with a CHC motivated them to pursue a career to support other children with a CHC, such as healthcare professions and research about a CHC. Siblings would bring their personal experiences about a CHC into their professions, such as an understanding about a CHC in research or how to interact with families. Their personal experiences about a CHC also motivated them to use their academic knowledge to create resources, such as mobile applications or tools that children with a CHC could use. Both parents and siblings identified multiple roles that siblings had in the family. Siblings continued to maintain a close relationship, and when one sibling had an acquired CHC, the siblings would learn how to provide support to each other. For example, a sibling described how he went to therapy appointments with his brother who had an acquired brain injury. The sibling provided both support and humor by being present at the therapy appointments, and the parents described how the sibling became a part of the care team. Adult siblings described challenges that they had when they became a caregiver, such as the sacrifices that they had to make with living with their sibling with a CHC or not being able to work full-time.

Emotional Outlet and Support for Siblings

Siblings wrote in blogs that they shared with the community about both the positive aspects of their relationship with their sibling with a CHC as well as the challenges. Siblings shared the message about how they are not alone, where their thoughts and feelings matter. Some siblings pursued their own goals and happiness, and also chose their roles with their sibling with a CHC. Some adult siblings experienced guilt when they did not voice their opinions. One adult sibling shared her sense of guilt when her brother was sent to an institution that the family believed was a good option at the time. Siblings spoke about how their emotions were connected to the emotions that their parents were experiencing, such as frustrations and stresses. Some siblings wanted to find ways to address the challenges such as learning how to help their sibling with a CHC. With the different emotions that siblings were feeling, they sought ways to have an outlet to express their emotions. A Photovoice program was offered to siblings and young patients with cancer, where they took photographs that represented their experiences that were later displayed at an event for the public community. Siblings were developing skills in how to cope with their emotions, and parents identified how these siblings would develop personal skills such as being caring and empathetic. Siblings also shared about the importance of open communication with parents because siblings might have hidden emotions. Siblings identified that they might not initiate discussions with parents about their feelings, and parents should create a space for these discussions.

Advocacy to Raise Awareness About Disability and Role of Siblings

Some siblings became advocates, in which they expressed a need to explain what disability was to their peers. For example, a sibling of a brother with autism described how she read a book to her class to explain autism and she often took the time to answer questions from her classmates. Siblings valued the connection that they had with other siblings who had similar experiences. Some siblings grew up without knowing about other siblings who have a sibling with a CHC. Siblings wanted to connect to a community of siblings to not only advocate for CHC and disability awareness and supports, but also learn about the role that siblings can have. For example, some siblings did not realize that they developed skills that could be well-suited for healthcare professions. One sibling described how she learned about the profession of a child health specialist after connecting with another sibling. Furthermore, adult siblings who were caregivers or guardians of their sibling with a CHC they identified how their roles were often not recognized at work. For example, employers recognized when co-workers needed to leave to take care of their child but not for their sibling with a CHC. Siblings identified how there should be recognition of the important role that they have. They all wanted to be part of a community where they can create change and advocate for a diverse community that their own sibling with a CHC could meaningfully participate in.

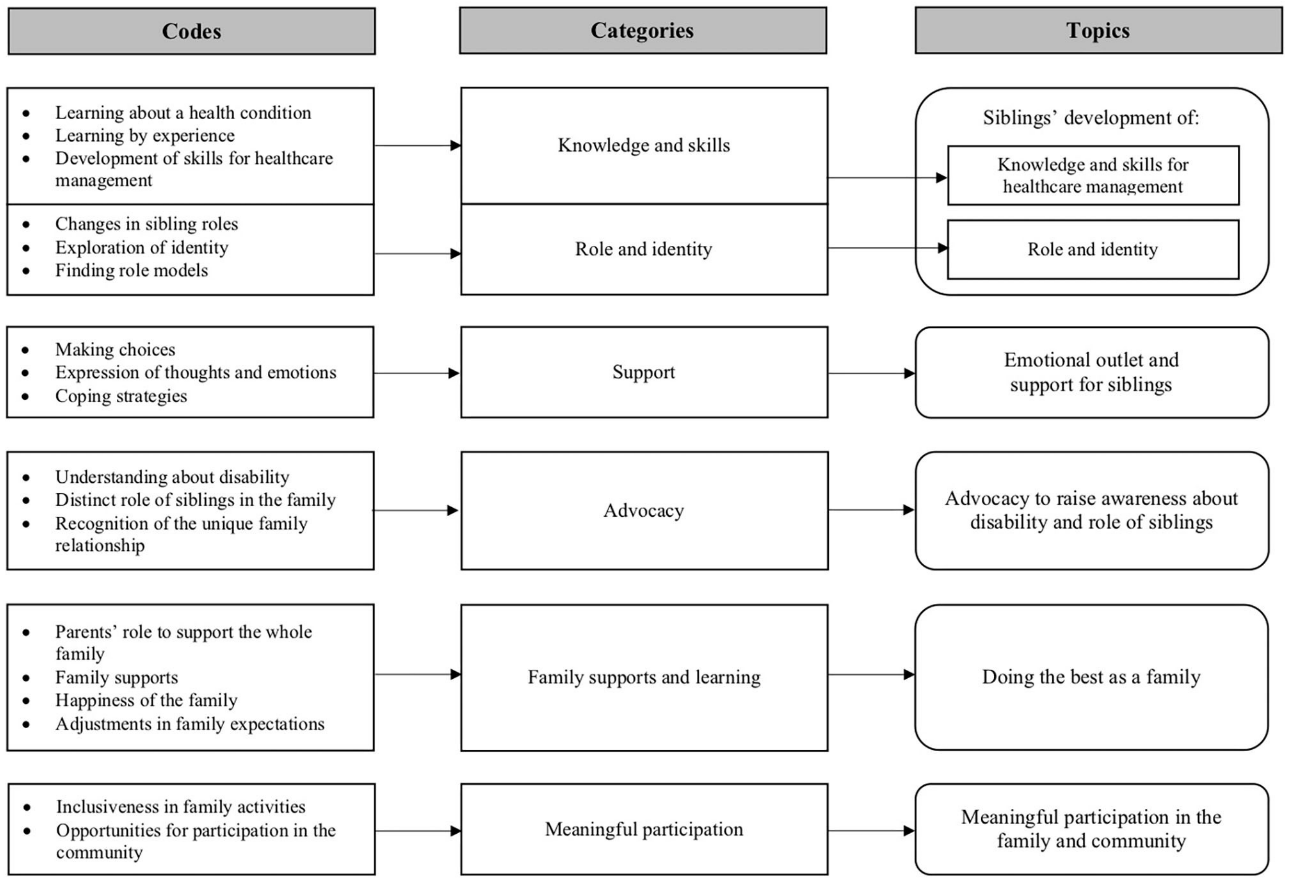

FIGURE 2 | Codes, categories, and topics indentified the blogs and interviews.

Meaningful Participation in the Family and Community

Families identified the importance of creating an inclusive environment where a child with a CHC can participate in activities. Some families planned trips and made sure that they rented adaptive equipment to ensure that their child with a CHC could participate in activities, such as hiking, biking, or kayaking. In their daily lives, young siblings shared in their blogs that they made sure that their sibling with a CHC was included in the games that they played with their friends. One family thought about different ways that every member of the family could participate in activities. When a sibling might be attending speech therapy, other family members could coordinate to have the other siblings participate in a sports activity at the same time. Parents identified how it was important to make sure that all siblings could meaningfully participate in the community. Both parents and adult siblings expressed their concerns about opportunities for their sibling with a CHC to participate in the community in the future. Siblings shared the positive value of a job for their sibling with a CHC, which provided a sense of pride to participate in the community. Some siblings wanted to address concerns about how to create an inclusive community for people with CHCs, and they created mobile applications to encourage their sibling with a CHC to develop the skills needed to participate in the community. For example, one sibling created a mobile application with a set of cards with which an individual with autism could practice the skills they needed to carry out an activity, such as taking public transportation. Both parents and siblings sought opportunities for a sibling with a CHC to participate in the community as they grow older.

Doing the Best as a Family

During separate interviews, parents shared about how they were doing the best that they could as a family and siblings shared how the journey of every family was different. A mother shared in her blog about experiences with raising her children, including children with a learning disability and Down syndrome, and she needed to time to learn about her children. For other parents, they learned about the different types of supports that would be appropriate for their child with a CHC and there was no "one size fits all" approach. Some parents initially chose to keep their life private, and they did not want to burden others with the responsibilities in caring for their child with a CHC that they feel were their own. They gradually recognized how it was important to reach out to others for support, such as their children and neighbors. Some parents also sought respite services to take care of their own health in order to optimize the care that they could provide to all of their children. In addition to services,

TABLE 2 | Frequency of topics identified in interviews and blogs with parents and siblings.

Topics		Siblings		Parents	
		Blogs (*n* = 18)	**Interviews (*n* = 26)**	**Blogs (*n* = 13)**	**Interviews (*n* = 12)**
Siblings' development of:	a) knowledge and skills for healthcare management	7	8	4	5
	b) role and identity	12	16	4	6
Emotional outlet and support for siblings		7	12	–	3
Advocacy to raise awareness about disability and role of siblings		6	3	–	–
Doing the best as a family		–	2	5	6
Meaningful participation in the family and community		6	4	–	5

–Topic was not identified.
** Multiple topics may be identified in each interview and blog.*

parents described the value of building a network of supports, such as connecting with other families with similar experiences. They wanted to have opportunities to meet other families and participate in activities that included the whole family. Some families created videos and films to share their story of both the positive experiences and challenges with other families.

SibYAC Reflections on the Findings

After synthesizing the findings from this review, the SibYAC members were asked to share their perspectives about the meaning of these findings. There were key topics raised in the blogs and interviews included in this review, and the SibYAC members were asked about whether these topics resonated with their own experiences of siblings of individuals with a disability. Siblings who wrote the blogs and inteviews included in this review identified that they wrote blogs as a way to share their stories so that other siblings would know that they are not alone, and writing blogs was an outlet for their emotions. Similarly, SibYAC members also wrote personal blogs about their personal experiences as a sibling and the roles that they have had. One SibYAC member shared an excerpt of her journal while her brother, who has cerebral palsy, was in a rehabilitation hospital after orthopedic surgery: "As my brother began to see progress into the next day, so did I. As he found a rhythm and learned the shuffles of the hallway, so did I. And before I knew it, I fell head over heels into the routine of physical and psychological exhaustion but unimaginable emotional fulfillment." She shares that her personal experience is a clear example of why consciously integrating siblings into the family-centered care model is so important.

While the findings of this review help to identify key resources for general information and information of how siblings can provide support with healthcare management, the SibYAC continued to identify that there is a need for advocacy to raise awareness about the important roles that siblings have. They often had to learn to develop knowledge and skills, in order to have a role with supporting their sibling with a disability with healthcare management. A SibYAC member shared: "There is no handbook for special needs siblings. It's not something that's majorly talked about and kind of always felt like a big secret. Every day, I am learning more about how to appropriately support my sibling through the transition from pediatric into adult healthcare." While this review identified that there are resources available for siblings of individuals with a CHC, few resources offer support for how siblings can be involved with the healthcare management of their sibling with a CHC.

As the SibYAC reflected on these findings, there is a critical gap in which there are no online resources available from Children's Healthcare Canada to support siblings in conversations about healthcare management and future planning to their sibling with a CHC, even though many siblings might already be part of the care team. A SibYAC member shared her personal experiences with this gap: "There has never been planning about the present, day-to-day things, let alone future planning, that has included me. The extent that I have been involved with my siblings is equal to the amount of intention and force I used to create a space for myself." The SibYAC shared how discussions about future planning can be helpful for families to ensure that there is clear communication about the role of siblings.

DISCUSSION

This review identified a variety of resources and documents available in English for siblings and families of children with CHCs across organizations of Children's Healthcare Canada. Most resources consisted of general information for siblings and families: to become aware and learn about different CHCs through books, news articles, and podcasts. There is an increasing trend in the use of the Internet for health information among patients, their families and general public (113), and each family requires different types of information based on their needs (114). In a qualitative study to explore the experiences of parents of children with disabilities who sought information, parents used online information to supplement the information provided by professionals (38). When healthcare professionals did not provide enough information during a consultation, some parents searched on websites of hospitals and rehabilitation centers for additional medical information (38). In the search for information, parents may also identify resources for how they can support the siblings of a child with a CHC. This review retrieved booklets for parents about how to communicate with siblings of children of individuals with a CHC.

In this review, few resources were identified to support siblings in the role of healthcare management with their sibling with a CHC. Similar to parents, siblings might also have questions and would like to have more information to support their roles in the healthcare management of their sibling with a CHC. Siblings of individuals with a CHC require skills and knowledge if they choose to have an active role in the healthcare management their sibling with a CHC. There are tip sheets that provided guidance for siblings to build a relationship with their sibling with a CHC, such as playing games or doing activities, as well as how to be a part of the care team (111, 112). There is also a toolkit that originally developed for siblings of individuals with an acquired brain injury, recently expanded to include different disabilities (105). It is important to consider and tailor different resources to prepare for the roles that siblings might choose to have, including with healthcare management of their sibling with a CHC.

This review highlighted a key gap in the needs of siblings based on the personal stories that they shared through blogs; many identified a need for emotional support. Blogs can be helpful for siblings, where they might find comfort to know that they are not alone (115). Siblings require acknowledgment of their emotions, and for some siblings, they are learning how to address their emotions as they continue to have a role in healthcare to their sibling with a CHC. Siblings might choose to seek online support to be part of a community with others who have similar experiences (115). Parents and families have previously described the importance of being a part of an online community where they can seek resources and connect with other families (38, 116), and siblings might have similar motivations to connect with other siblings online.

Siblings also shared, in blogs and interviews, their perspectives about the importance of advocacy to raise awareness about CHCs and their own roles. Siblings may need to advocate for their role in the family. This review identified that siblings require additional resources in order to learn and be prepared for their future roles. The extent of discussions about future planning can vary in families, and siblings are often not included in these discussions (13, 117). While some parents may wish for the siblings to have their own separate lives, siblings shared in qualitative studies that they chose to have active roles such as being a caregiver and they identified the need to have conversations with their parents about future planning (118, 119). Discussions about future planning can be helpful for families, providing an opportunity for siblings to identify new or changing roles and to facilitate the sharing of information between parents and siblings (31, 33). These discussions can be ongoing to adapt to the changing situations of the family over time. While these discussions can be challenging and complex for families, siblings identified the need to have clear plans so that they can be prepared for their future roles (117, 119, 120).

Many families shared how they are learning from experience and doing the best that they can with their family member with a CHC. In this review, both parents and siblings described the positive value of creating a supportive and inclusive environment of a person with a CHC. This inclusive environment applied to the family environment where some families sought opportunities to participate in different activities, as well as in the community such as having a job. Both parents and siblings described the concern that they had for an individual with a CHC after graduating from high school, and they were worried that there may be fewer opportunities to participate in the community. This concern about the transition to adulthood for individuals with a CHC has been raised in the literature (121, 122), and current news noting that there are ∼12,000 young and middle-age adults with disabilities in Ontario, Canada who are on a waiting list to seek supports and residential care (123). In addition to employment opportunities, both parents and siblings have future worries for their child with a CHC such as the navigation from pediatric to adult healthcare services (122, 124). Some siblings shared in blogs and interviews how they gradually learned to take on caregiving responsibilities. Siblings are becoming adults and they may take on future caregiving responsibilities. They are often learning through experience about how to care for their sibling with a CHC throughout the lifespan (119).

Strengths and Limitations

A strength of this review is the involvement of the SibYAC as advisors throughout the process of this review. They provided their perspectives on the aims of the review, data analysis of resources, and future directions on how to disseminate the findings and develop future resources. Another strength of this review is that the resources identified have been compiled and can be applied to enhance existing resources for siblings of individuals with a CHC and inform the co-development of future resources. A limitation of this review is that the information that can be extracted from the documents included may be restricted by the purpose of the document and the content that the creators choose to share. Another limitation is that all documents were identified from children's hospitals and treatment centers that are part of Children's Healthcare Canada were in English and excluded documents in French. The documents and resources from the websites of organizations that are part of Children's Healthcare Canada might be selectively published and might not include information about the care that these organizations provide. The information may not be reflective of the entire landscape of resources for Canadian siblings of individuals with a CHC. In addition, at the time of this review, documents and resources were retrieved from 31 organizations that were a part of Children's Healthcare Canada. Since then, eight additional organizations have been included. Most organizations that are part of Children's Healthcare Canada are children's hospitals and rehabilitation centers, and resources from services offered in the community, such as mental health services, might have been missed. However, the resources and documents in this review provided a starting point for identifying general information and information about how siblings can support their sibling with healthcare management. Additionally, while data extraction and coding was conducted by a single analyst which limits our ability to report on inter-coder reliability, the categories and meaningful clusters of data that were developed were reviewed and discussed by two key stakeholder groups, a form of analyst triangulation and peer debriefing that enhances overall data credibility.

Future Directions

This review highlighted key gaps that can be addressed in the future in order to optimize supports for siblings of individuals with a CHC. First, access to existing resources for siblings should be improved by compiling and storing them in one place. As knowledge translation and dissemination can include multiple strategies, there can be multiple formats of resources created, such as infographics, toolkits, videos or podcasts. The Health Hub in Transition in Canada (125) and the F-words for Child Development Knowledge Hub (126) are examples of where information and tools are available online. The uptake and impact of the knowledge translation and dissemination strategies should be evaluated. Second, there should be resources to support siblings in the healthcare management of their sibling with a CHC. In this review, both parents and siblings shared in blogs and interviews the important role of siblings in healthcare management. Despite the important role that siblings might want to have with healthcare management, there are few resources available to support and empower siblings in this role. Third, this review identified that there are no resources in English available within the online materials from the organizations through Children's Healthcare Canada for parents or siblings to facilitate ongoing conversations about the roles that siblings would like to have with their sibling with a CHC. The conversations could also include the topic of healthcare transition about how youth, siblings, families, and healthcare professionals can help youth prepare for the transfer to adult healthcare (127). Tools could be developed to facilitate these discussions in the family and with healthcare professionals (32, 118). Finally, resources could be developed for other professionals, including teachers and healthcare providers, to encourage discussions about the experiences and roles of siblings beyond healthcare management. Siblings have identified that they wanted more information about future responsibilities, such as legal and financial information regarding the care of their sibling with a CHC (22, 24).

CONCLUSION

This review identified resources for siblings that are available from children's hospitals and organizations that are part of Children's Healthcare Canada. Resources that are available for siblings of individuals with a CHC mainly address general information, such as support programs and workshops. There are some resources, such as tip sheets and a toolkit, to offer strategies for siblings to learn about the healthcare management of their sibling with a CHC but these resources are only available at two children's hospitals. Siblings shared about their experiences in blogs and interviews, including their development of knowledge and skills for healthcare management, as well as roles and identity that often relate to the healthcare management of their sibling with a CHC. There is a key gap in available resources, in which siblings and parents identified that knowledge and skills for healthcare management is an important role for siblings but there are few resources that provide this information. Given the needs expressed by siblings, future resources should be developed to share information about healthcare management for siblings, as well as tools to facilitate family discussions about the roles that siblings would like to have in the future. A synthesis of the identified resources could be shared in an accessible format, such as in an online hub, for siblings and families.

AUTHOR CONTRIBUTIONS

LN, BDR, SJ, MK, and JWG contributed to the conceptualization and design of this review, and perspectives from HD, SB, and JH informed the review aims. LN drafted the manuscript. HD, SB, and JH contributed to the draft of the Reflections section. All authors provided input for the analysis, interpreted the findings, revised and reviewed the manuscript, and provided their final approval.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.724589/full#supplementary-material

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A Mixed-Methods Feasibility Study of Integrated Pediatric Complex Care: Experiences of Parents With Care and the Value of Parent Engagement in Research

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Introduction: Children with medical complexity (CMC) are among the most vulnerable children in society. These children and their families face challenges of fragmented care and are at risk for poorer health outcomes. Families with CMC play a vital role in providing care and navigating the complexities of healthcare systems. It is essential to understand the best ways to engage these families in research to improve the care and optimize the health of CMC.

Objectives: This study explored parent engagement within the context of a feasibility study evaluating an Integrated Tertiary Complex Care (ITCC) clinic created to support CMC closer to home. This paper aimed: (1) to understand the family experiences of care and (2) to explore parent engagement in the study.

Method: This mixed-methods feasibility study included three components. First, feedback from focus groups was used to identify the common themes that informed interviews with parents. Second, one-on-one interviews were conducted with parents to explore their experience with care, such as the ITCC clinic, using an interpretative description approach. Third, the questionnaires were completed by parents at baseline and 6-months post-baseline. These questionnaires included demographic and cost information and three validated scales designed to measure the caregiver strain, family-centered care, and parental health. The recruitment rate, percentage completion of the questionnaires, and open-ended comments were used to assess parent engagement in the study.

Results: The focus groups involved 24 parents, of which 19 (14 women, five men) provided comments. The findings identified the importance of Complex Care Team (CC Team) accessibility, local access, and family-centered approach to care. The challenges noted were access to homecare nursing, fatigue, and lack of respite affecting caregiver well-being. In this study, 17 parents participated in one-on-one interviews. The identified themes relevant to care experience were proximity, continuity, and coordination of care.

The parents who received care through the ITCC clinic appreciated receiving care closer to home. The baseline questionnaires were completed by 44 of 77 (57%) eligible parents. Only 24 (31%) completed the 6-month questionnaire. The challenges with study recruitment and follow-up were identified.

Conclusion: Family engagement was a challenging yet necessary endeavor to understand how to tailor the healthcare to meet the complex needs of families caring for CMC.

Keywords: qualitative, mixed methods, feasibility study, children medical complexity, complex care, patient engagement

INTRODUCTION

Children with medical complexity (CMC) are among the most vulnerable children in society. They have multiple chronic conditions and significant functional limitations and are dependent on life-sustaining technology, such as tracheostomy, home mechanical ventilation, and enteral feeding tubes for daily survival (1–3). CMC are at very high risk of multiple and prolonged hospitalizations, frequent medical errors, and poor health outcomes (4, 5). CMC comprise only 0.67% of all children in Ontario (6), but this group utilizes about one-third of all the provincial child health resources, thereby having a substantial impact on the healthcare system (1, 7, 8). A study from the Canadian Institute for Health Information reported that over a 2-year period, the hospital costs associated with children and youth with medical complexity were $866 million (9). CMC accounted for 37% of all hospital admissions and 54% of total days in the hospital (9). Similarly, the impact of caring and coordinating complex, fragmented care for such children is substantial for the families.

Care for CMC requires close monitoring by multiple healthcare providers (10–12). Different care delivery models exist for CMC, such as primary care-centered frameworks, consultative or co-management-centered models, and episode-based models (10). The primary care-centered frameworks can be based in the community or tertiary care centers and focus on providing coordinated care for CMC through a dedicated primary care center (10). These models, where offered, serve as the first entry-point for CMC to access the healthcare (10). The co-management-centered models, on the contrary, are not the first entry-point for CMC and encompass subspecialty providers in tertiary centers coordinating care with primary care providers in the community (10). Last, episode-based models focus on providing care during a discrete and acute period of illness and are usually time-limited in nature (10). Examples of episode-based models include transitional care homes where CMC are between hospitalization and home. The successful complex care programs at tertiary care centers deliver better care at a lower cost due to a reduction in preventable inpatient and emergency department visits (12–15).

Despite the benefits of different models of care, CMC and their families still face barriers when accessing the complex care programs in tertiary centers. Since many children do not live near the tertiary care centers (16), traveling to these centers can present financial, physical, and social challenges for the caregivers and their CMC. Further, poor communication across the healthcare settings may limit the appreciation of tertiary healthcare providers of the breadth of community services that can provide additional support to CMC (10, 17). The strategies to overcome these barriers include creating enhanced primary care center-based complex care programs dedicated to care for CMC with resources and staff centralized at a tertiary center, care coordinators, and standardized care coordination quality improvement tools (1, 10, 14, 15, 18, 19). Integration of a tertiary care center with a community-oriented pediatrics team, different from the previous CMC care models, has been shown to provide cost-saving benefits, increased family-centeredness, decreased hospitalization rates, decreased parental work loss, and higher family and healthcare provider satisfaction (20–22).

The Integrated Tertiary Complex Care (ITCC) clinic is the first clinic of its kind where a tertiary complex care clinic is embedded in a treatment center of children. The ITCC clinic is located within the Niagara Children's Center, 80 km from the McMaster Children's Hospital (MCH), a large tertiary hospital for children (23). Established in 2015, the ITCC clinic is a collaboration between a tertiary academic hospital and regional children's treatment center, created to coordinate, support, and bring care closer to home for CMC. Implemented as a monthly full-day clinic, the MCH CC Team consisting of a pediatrician, nurse practitioner, and respiratory therapist travel to Niagara to conduct clinics in partnership with the allied health and community team at Niagara Children's Center (24). The aim of the ITCC clinic is to provide comprehensive, holistic care for CMC, improving communication between the tertiary, community healthcare partners, and parents of CMC while alleviating the travel burden to a tertiary center. It is important to evaluate the ITCC in comparison with the existing models of care to ensure CMC are receiving optimal care.

To evaluate the ITCC clinic, parent engagement is necessary to capture the perspective of the user of this system. Parent engagement can include consulting, providing information to inform decisions, sharing leadership, and defining agendas (25). It is notable that the higher levels of engagement, such as shared leadership, are not always desired by the patients and families, and not always the most effective, depending on the research and clinical context (25). Increasingly, the researchers and clinicians view parent engagement as an essential way to inform better care and include patient experiences to help balance an unstated bias toward the clinical and system outcomes (25). A recent literature examining the quality of care for CMC has emphasized the need for parent research involvement through consultation to evaluate the implemented improvements in care provision and provide crucial feedback to the providers to facilitate the sustainable changes (26).

A pilot study was conducted to evaluate the ITCC clinic model of care for CMC, reported in a forthcoming paper. This paper outlines the findings of a secondary data analysis of the pilot study, aiming: (1) to understand family experiences of care and (2) to explore parent engagement.

METHODS

This mixed-methods feasibility study involved a Family Engagement Day with focus groups, one-on-one interviews with parents, and the completion of questionnaires. The study took place from October 2016 to March 2018 at MCH and the newly created ITCC clinic within Niagara Children's Center in the Niagara Peninsula. Ethics approval for all aspects of this study was obtained from the Hamilton Integrated Research Ethics Board prior to the recruitment (#1011).

Participants

The parents of CMC, from two different models of care, within the catchment area of MCH who were followed by the CC Team were recruited for the study. The parents who met the study eligibility criteria and consented to be contacted by the research team were invited to participate. They were subsequently screened for eligibility with the following criteria: they were the primary caregiver(s) to the patient and they could read and understand English. The parents were excluded from the pilot study for reasons that included caregiver hardship (such as the parent being ill or child being acutely ill). An informed written consent was obtained from all the participants through clinic visits or by mail.

Data Collection

This study is a secondary analysis using data collected as part of a pilot feasibility study evaluating an ITCC model of care. It involved three sources: focus groups, one-on-one interviews, and questionnaires. The data collection methods used in the feasibility study are described below.

Family Engagement Day

As part of a Family Engagement Day held in March 2016 at MCH, focus group discussions were facilitated by the CC Team. The focus groups were designed to engage parents and healthcare providers to identify the challenges and facilitators with the goal of improving the delivery of care to CMC. The day involved 24 parents, 24 healthcare providers, and 12 stakeholders (e.g., managers of Local Health Integration Unit (LHIN), CEOs, and school board representatives). There were five breakout groups, each consisting of a facilitator (member of the CC team), parents (ITCC and tertiary care), a healthcare provider, and a stakeholder. The breakout groups were used during the two sessions. In the first session, "Understanding Your Needs – Value Stream Mapping Exercise," the participants were asked the following: (1) Describe your ideal care and current challenges; (2) What does a typical care journey look like?; and (3) Describe how you feel during your care journey. During the second session, "What Matters Most? Interactive Discussion," the participants were asked to discuss the key elements for successful complex care and coordination, such as: Medical Care Coordination, Home and Community Care Coordination, Knowledge Building for Families, and Community/Partner Engagement. The key findings in the feasibility study included the needs of families: (1) better communication between inpatient and outpatient services, between agencies, and between the hospital and community services, (2) coordination of multiple appointments, and (3) trained professionals that are competent and willing to care for a child with medical complexity. These findings were used to structure the questions in the interview guide for the next stage of the study. Focus group sessions were audiotaped and transcribed. The manual identification of themes was done by two coders using a line-by-line approach as a unit of analysis.

One-On-One Interviews

Recruitment for one-on-one interviews took place from August to December 2017. A purposeful sample of parents was recruited from tertiary hospital catchment regions with a focused representation of parents from the Niagara region. A written consent was obtained at the time of the interview or through mail. Informed by the focus group findings, the aim of the interviews was to gain an in-depth understanding of the family experiences of care at both the tertiary care site and the ITCC clinic, as well as care coordination through the new model of care at the ITCC clinic.

The interviews were conducted by an experienced qualitative researcher who used a semi-structured interview guide. After the initial eight interviews were completed, an interim analysis was conducted to further refine the interview guide. The interviews were transcribed, de-identified, and analyzed using NVivo 10 software. The interviews were coded line-by-line to identify the themes and subthemes relating to family experiences of care and parent engagement. The interviews were conducted until no new concepts were identified. Prior to analysis, the codes and full interview transcripts were reviewed by OH to ensure that coding accurately captured information in the full transcripts. Informed by the interpretive description method, the interviews were analyzed using a rigorous constant comparative and iterative approach to identify and describe the themes and subthemes (27). The results were then shared with the team, discussed, and refined.

Questionnaires

During the feasibility study, the parents were invited by phone or in person to complete the questionnaire online or using a paper booklet. Timing of completion was either during a clinic on the day of scheduled appointment of their child with the CC Team, or outside the clinic at the time convenient for the family. All data were entered into a Research Electronic Data Capture (REDCap, Vanderbilt University, TN, USA) database supported by the Department of Pediatrics at McMaster University, ON, Canada. A link to complete the 6-month follow-up questionnaire directly into REDCap was sent *via* email to the parents who provided an email. Those who did not provide an email were given a paper copy of the questionnaire in the clinic.

The questionnaire included demographic questions, collected through standardized survey questions, such as caregiver age, gender, race, marital status, relationship to child, and education. The parents were also asked about their health and medical condition of their child. The impact of the chronic illness of the child on their family was measured using the Impact on Family Scale, a 15-item scale that notably examines the financial burdens as well as emotional concerns for families (28). Parental perceptions of whether the care provided displayed family-centeredness were assessed using the Measure of Process of Care (MPOC-20). The MPOC-20 is comprised of five subscales: respectful and supportive care, enabling and partnership, providing general information, providing specific information, and coordination and comprehensive care (29). The EQ5D-5L (EuroQol Office, The Netherlands) was used to assess the health status of the caregivers. This scale asks about five dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression (30). A series of questions focused on out-of-pocket costs for medical care were included to assess how well this data could be obtained from the parents. Finally, at the end of the questionnaire, an open space was available for parents to provide any additional comments they had for the research team. The open-ended comments were coded to identify the themes and subthemes relating to family experiences of care and parent engagement.

Parent engagement was further examined in questionnaire data by assessing both the response rates to the questionnaires as well as completion rates of each section of the questionnaire.

TABLE 1 | Demographic characteristics of participants in one-on-one interviews (*N* = 17).

Characteristic	*N*
Gender	
Female	13
Male	4
Age	
20–29	2
30–39	7
40–49	6
Missing	2
Race	
White	15
Other	2
Marital status	
Common law/Married	13
Single/Separated	4
Relationship to child	
Biological Mother	12
Biological Father	4
Foster Parent	1
Education	
University or College Degree/Diploma	12
Some Post-Secondary (University or College)	3
Missing data	2
Employment status	
Employed full-time	9
Employed part-time	4
Receiving social assistance	2
Receiving unemployment insurance	1
Student	1

RESULTS

Aim #1: Family Experience With Care

This section draws on the focus groups, one-on-one interviews, and open-ended comments from the questionnaires.

Focus Group and One-On-One Interviews

The focus groups involved 24 parents, 19 (14 women, five men) of which provided comments during the session. The analysis of the focus group sessions identified the importance of available tertiary care providers, a coordinated care plan, local access to care, and a family-centered approach to care delivery. The findings included the challenges related to accessing trained community nursing, social isolation, and lack of respite, all of which contributed to the burden of care and adversely affected the well-being of caregivers. The participants welcomed and valued the integration of complex care services into the community.

One-on-one interviews were conducted with 17 parents (as shown in **Table 1**). The key themes from the interviews illustrate family experiences related to the following: proximity and continuity of care closer to home, care coordination, accessibility and communication, and family-centered care. Themes 1 and 2 are specific to the ITCC clinic while the continuity and coordination of care were also reported to be valuable to the partcipants receiving their care at the MCH.

Theme 1: Complex Care—Proximity and Continuity of Care Closer to Home

This theme builds on the findings from the Family Engagement Day focus groups that highlighted the challenges associated with disjointed care, travel, and long-distance appointments. In the one-on-one interviews, the parents whose child received care through the ITCC clinic appreciated the proximity of care to their home and the easier access to the allied health professionals. Receiving care closer to home was reported to have many advantages. The parents appreciated that, in addition to being close, the team came well prepared with the medical supplies and had access to the medical records of their child:

> *I think what works for me is that it's close and that it's Dr. [name]. And they . . . come equipped with quite a few things. So, last time . . . normal saline. . . nebules [were] on back order, and . . . they had some. And they had silver nitrate . . . that they were able to give us [for granulation tissue]. So, they come well prepared. . . . And it's nice that they have access to [child's] lab values. . . so we were able to*

> *bring that up … I just love that it's close... It's still 40 min [drive], but it's not an hour and a half, so it's less than a couple hours in the car.* (P1)

Since the proximity of the clinic meant shorter travel times to the appointments, the parents reported easier logistics, requiring fewer arrangements regarding the time taken off work, childcare for other children, packing all the necessary medical equipment, and navigating unfamiliar hospital and parking environments. Having appointments in the community was less disruptive to the lives and schedules of families:

> *So, for [child] to go to [the tertiary center], [they miss] a whole day of school… and [they're] already behind, so missing a lot of days, it impacts [them] a lot. … If [child] just has to go up the road… then [they] only miss half a day, if that.* (P16)

Receiving care through the Children's center also meant a unique benefit for children attending the preschool on site. The parents appreciated the convenient access to the allied health professionals at the center, with some parents wishing for an extension of these services for school-aged children as well.

In addition to proximity, the parents emphasized that familiarity of the team with their child made the process of receiving care less overwhelming for the family:

> *I love that it's Dr [name]. [With another physician] … I'm having to retell [child's] story, and they don't know [child] … If it's any other physician, then I will ask … to schedule me in when Dr [name] is going to be in Niagara. … I love that it is a team … and … the same nurse practitioner every time.* (P17)

> *I know that group of people well…. [the nurse and respiratory therapist]… know the kids well … [and] … all the pertinent questions to ask. It's an easy… process. Easy to remember everything you need to talk about and you get things done.* (P1)

With proximity and continuity came important relationships that allowed for more proactive and timely care, sometimes eliminating the need to go to the Emergency Department (ED) or Emergency Room (ER):

> *And just being able to say, "This is what's going on. What do you think?" And for [the physician] to say, "You need to go to the ER or we're running a clinic today. Why don't you come in to the clinic? And we've been able to treat [the sick child] that way. So… having the close relationship and the continuity… is pretty important.* (P1)

Some parents suggested increasing the number of clinic days at the Niagara Children's Center and emphasized the need for access to the trained staff on a regular basis to meet extensive and often unexpected medical needs of their child:

> *There's a couple of times [when the child had an episode] … and it's just we've been fortunate enough that it's been a Complex Care day … and the team has been down here. So, we were able just to drive the 7 min to it…* (P14)

Given the complexities of their children, parents often emphasized the vital importance of continuity in their care:

> *[Child]'s very complex, and unless you see [them] often… And I know… we are one of very many patients part of the Complex Care team. So, it's really hard to invest in your patients when you see them a few times a year... With that said, I think that we get excellent care.* (P17)

The continuity of care, consistency, and expertise were valued more highly than proximity of the clinic to their home, so that some families would still be prepared to take the drive to the tertiary center to be seen by the CC Team familiar with the medical complexity of their child.

Theme 2: Care Coordination

Having experienced fragmented care, the parents overwhelmingly expressed their appreciation for care coordination through the CC Team. A complex care system navigator scheduling and coordinating appointments with various subspecialty clinics made a significant difference: It minimized travel to the hospital and distress to children and their family.

Given the systemic challenges parents often face with care coordination, the parents attending the ITCC clinic appreciated the simplified process and the effort of team to coordinate and streamline the complex care of the child despite the inherent challenges:

> *They try to make things easier when they can. Dr. [name] is always pushing to try and get appointments together. [Doctor] would like to see me not have to travel as much.* (P1)

Care coordination by the CC Team reduced the stress in very important ways. For example, having all involved healthcare providers communicate and synchronize their plans at the ITCC clinic provided a comprehensive holistic approach to care and was reported to be very helpful to families:

> *They can… share information with therapists at the Children's Center as well. … Any issue, they can speak with each other about it … to come up with a plan. And it's not me having to go and try and find things out… get information from this person here, and then get information from this person there. They're all linked together. So, everybody is in sync. It's less stressful.* (P16)

Care coordination was often described as easier to manage, less time intensive, and less anxiety-provoking.

Theme 3: Accessibility and Communication

All parents agreed that different communication options, such as phone, text, or email made the CC Team members more accessible. In the group followed at the tertiary hospital, parents described access to the CC Team as excellent and the team as very responsive and accommodating, often allowing for parents to receive the clinic appointments when needed and allowing for same-day scheduling. Access to the team reduced the ED visits and other urgent care appointments for parents in both the tertiary hospital and the ITCC groups. Being able to reach the team over the phone often allowed parents to get faster care, or to determine if travel to the hospital was indeed necessary. Some parents valued the opportunity to send pictures and get professional guidance with advice that allowed them to manage the situation on their own. Access to the team minimized the need to travel, and reduced the stress and healthcare resource use for parents:

> *If I run out of a prescription or something wasn't [available]… Sometimes… I'll call to reorder a med, and they just don't send it. … The names on the bottle is often different doctors, right, and then it doesn't get to that same doctor. So, I know that I can always text and … it gets taken care of within 24 h. … So, that's extremely helpful. That takes a lot of stress and pressure off… otherwise it would be visit to come in and get a new prescription, 'cause you … can't go without it.* (P1)

Understandably, since many parents described travel with their child as stressful, costly, and emotionally exhausting, the parents valued the ability to access care virtually whenever it was possible.

Theme 4: Family-Centered Care

All parents agreed that the CC Team provided family-centered care and appreciated the relationships they had with the CC Team. For many parents, being validated and treated by the team as partners in the care and decision-making of their child was very important:

> *It's more… personalized. And you feel…included and … valued and you feel like, "I'm not just like a nobody here. I matter to them too. Or my opinion matters here too." … They do a really good [job] in making sure that you feel like part of the team.* (P16)

Personalized care also meant the CC Team attended to the social and educational needs of the child, and not only their medical needs. For example, the CC Team offering to do a school visit was not expected, but highly appreciated by one parent:

> *Right now I'm emailing back and forth with [doctor] to see if we can organize a school visit where … the team can come into [child's] class and answer… any questions students might have, and help in the social aspect … in school. So, even little things like that. … I would have never thought that that's a service that they provide… you only think it's just… medical. But they go far more than just the medical.* (P16)

The parents who attended the ITCC clinic described the team as responsive, respectful, and understanding, giving them time to ask questions and acknowledging that their child may present differently. Such understanding often translated into flexible thinking and treating each child as an individual first:

> *I never feel rushed. I always feel like I can ask as many questions as I want. I never feel … that they're watching the clock and they're waiting for the next patient to come in, which I really do appreciate. And I always feel that we are asked how things are going and what we think is going on, because [child] presents very differently. For an ear infection, [child] is not pulling at [child's] ear or rubbing it … [child] usually doesn't spike a temp. [child] is usually throwing up because it's causing [child's] gag reflex to be all out of whack. So, they respect our opinion.* (P17)

The positive relationships with physicians, nurses, and staff were frequently reported to be a very important aspect of family-centered care throughout all the interviews. The parents highly regarded the responsiveness and accessibility of the CC Team to the needs of their child:

> *The relationship with the doctors here is excellent. I can always rely on them… to get the kids what they need. … I can always call when I have a problem.* (P1)

The parents felt grateful and often emotional when describing the team members as "saint[s]" and "guardian angels" who are "passionate about the patients" and show that they value their child by "treating their child as gold":

> *Doctors and people involved is that … five percent of people within their field who are not only experts, but they are empathetic. They get it. They understand your life is not typical … I … feel really fortunate that a lot of the five percent people have come into my life. … I always say that if I could change any… everything for [child], I would in a heartbeat, but I would… [crying] I think I would feel lost because I've met so many great people because of [child].* (P17)

Open-Ended Responses From the Questionnaire

In total, 21 parents chose to provide open-ended comments *via* free form text at the end of the questionnaire.

In terms of family experience of care, most comments expressed appreciation of the parents for the care their children received through the CC Team. The team was described as "professional," "supportive," "caring," "kind," and "exceptional." The parents reported that they trusted the knowledge and expertise of the team about the care of their child and felt listened to and not rushed during their appointment. The parents were grateful for the help of the team with their concerns, coordination of appointments in an accommodating way, and how the team celebrated the milestones of their child' with enthusiasm. The parents appreciated how the team tried to make their lives easier and were understanding of their circumstances, which they noted was not the case outside of the complex care clinics. Overall, the parents highlighted that it was difficult advocating for their child on their own and they underscored the role of the CC Team beyond a "transactional" one. Some parents were grateful to the team for teaching them how to care for their child during very difficult times, whereas others attributed reduced hospital visits and the success of their child directly to the expert guidance of their CC Team. An example quote from a follow-up questionnaire shows the support from the perspective of a parent:

> *The complex care team is by far the most effective and competent team that we've dealt with over the past couple of years. You can tell they care and are invested in helping us make our child's health as best [as] it can be. They also work collectively to ensure there aren't gaps in our child's care. They go above and beyond to make sure we have what we need to care for our child and they give us hope*

TABLE 2 | The demographic characteristics of participants in the questionnaire study.

Characteristic		Assessment 1		Assessment 2	
Age (mean (SD))		37.3 (6.8)		36.7 (7.2)	
		N	**%**	***N***	**%**
Region	Hamilton or other	40	90.9	22	92
	Niagara	4	9.1	2	8
Gender	Female	39	88.6	19	79
	Male	5	11.4	5	21
Race	White	39	88.6	20	83
	Other	5	11.4	4	17
Relation to child	Biological Mother	35	79.5	19	79
	Biological Father	5	11.4	5	21
	Foster Parent	4	9.1	0	0

> *that this is what the health care system could look like if everyone invested the same time and energy into their patients.* (P46)

Aim #2: Parent Engagement in Research

Focus Group and One-On-One Interviews

In total, 24 parents of CMC were invited to attend the focus group, with 19 actively participating contributing comments in the transcripts. For the one-on-one interviews, 17 parents participated, with interviews lasting between 60 and 90 min.

In the one-on-one interviews, the parents viewed their engagement in research as validating their complex lives and as an indicator of family-centered care.

> *Doing these type of studies … of trying to figure out how it's affecting us. … It just goes to show how family-centered it really is.* (P16)

Questionnaires

In total, 49 (64%) of 77 eligible primary caregivers consented to complete a questionnaire. Of these, 44 (57%) completed the initial questionnaire and 24 (31%) completed the 6-month follow-up questionnaire. The initial sample included four parents of children who attended the ITCC clinic, of whom two also completed the follow-up survey. The demographic characteristics for the sample are shown in **Table 2**.

For the three survey tools that were administered, the sub-scale and overall scores were obtained for the majority of parents (≥92%), with the exception of two sub-scales of the MPOC. The MPOC sub-scales of "Providing General Information" and "Providing Specific Information," during the initial assessment, had slightly lower completion rates of 82 and 86%, respectively. **Table 3** provides summary statistics by assessment for the questionnaires.

To capture the cost-related data proved challenging. The completion of cost data ranged from 78 to 96% for parents reporting out-of-pocket costs. The costs were reported for visits to the emergency room, community pediatrician, and family doctor, as well as hospital admissions. The costs provided included transportation [bus, taxi, and personal vehicle (km)] and parking. The lowest completion rate at both assessment points was cost associated with visits to the community pediatricians. Prescription medication used by the child was reported by 34 and 18 parents at the initial assessment and follow-up, respectively. The complete cost data for both prescription and over-the-counter medication was reported as high (>90%) at both the initial and follow-up assessments. For devices and supplies at the initial assessment, 36 parents reported purchasing devices and supplies related to care, with nine having full coverage of costs by insurance. Of the 27 parents reporting out-of-pocket costs, 21 provided costs estimates. At the follow-up assessment, the completion rates for device and supply costs increased with 20 parents reporting this cost, and five reporting full insurance coverage. In total, 14 of 15 parents with out-of-pocket costs provided cost estimates with this variable. The overall costs could be calculated for all the questionnaires. However, only 32 of 44 and 18 of 24 parents at the initial and follow-up assessments, respectively, had complete cost data for all the elements. The remaining parents had at least one area of missing cost data, therefore the total cost value calculated might not be reflective of their experience.

Furthermore, the detailed open-ended comments from 21 parents provided insight into various areas of care as well as the questionnaire administration.

Challenges with the questionnaire completion identified by the parents related to time constraints and applicability of the questions to their family. The parents dedicated time to complete the questionnaire because they felt passionate about helping with the study and improving the care for the patient population. Some respondents indicated they would have preferred to engage in a conversation over the phone or in person, instead of completing a questionnaire. Feedback in the baseline questionnaires suggested that certain questions pertaining to costs needed to be "time specific" and to consider existing funding models:

> *If I was asked to complete this survey every year of [child's] life from birth to now, they would all look very different (far more medical appointments in [child's] first year–over 120).* (P41)
>
> *This survey would be easier and more accurate if you did a yearly overall expense of medical supplies and found out how much ADP [public] funding parents received...as opposed to individual items like catheters etc.* (P3)

Finally, the parents pointed out the unique circumstances of their family that made some questions not applicable (e.g., foster parents, parents who work full-time, and parents who work flexible hours). Some alluded to multiple complex factors in addition to the illness of their child's which "made some questions difficult to answer" and reminded the research team that "extraordinary expenses and circumstances do not fit in boxes but happen nonetheless." Some parents emphasized the often overlooked "opportunity cost" when the promised services and funding failed to deliver.

TABLE 3 | Summary statistics for the Measure of Process of Care (MPOC), EQ5D, and Impact on Family measures.

	Initial Assessment (N = 44)			Follow-up Assessment (N = 24)		
	Mean	**SD**	**% complete**	**Mean**	**SD**	**% complete**
MPOC						
Respectful & Supportive Care	5.7	1.1	95	6.1	1.0	96
Enabling & Partnership	5.6	1.1	95	6.0	1.1	96
Providing General Information	4.0	1.7	82	4.0	1.6	96
Providing Specific Information	4.6	1.8	86	5.0	1.7	96
Coordination & Comprehensive Care	5.5	1.2	95	5.9	1.2	96
EQ5D						
EQ5D Overall Score using CDN valuation	0.8	0.11	95	0.8	0.1	92
Impact on Family						
Impact on Family Burden Score	43.8	9.1	95	45.1	8.2	96
Impact on Family Financial Burden Score	14.4	4.0	95	14.4	3.0	96

DISCUSSION

This mixed-methods pilot study focused on the first integrated complex care clinic embedded in a children's treatment center that was implemented to provide multi-disciplinary complex care closer to home for CMC and their families. The ITCC clinic provided coordinated care for CMC and alleviated the burden of travel for families. Access to specialists trained to understand the comprehensive needs of their children in the community was an important aspect of the model of care to meet the continuous, intense needs of their children. Building on the knowledge gained from the Family Engagement Day focus groups, interviews, and questionnaires was used to understand family experiences of care and explore the parent engagement in the research study. Further, the data collected provided important information on the best ways to engage these families in research to improve care and optimize health of CMC. The parental perspectives provided valuable insights into the experience of their families with care, further demonstrating the importance of parent engagement in research (25, 26). The finding in our study regarding the parent capacity to fulfill the multitude of roles and tasks that go with caring and care coordinating for CMC is consistent with the other studies (31, 32). A recent systematic review suggests a growing body of research focusing on the health and well-being of CMC parents as primary caregivers (33). A scoping review on the interventions to improve the health and well-being of parents of children with special healthcare needs calls for careful tailoring to ensure that such interventions are both feasible for delivery within routine care settings, as well as relevant and accessible (34). The present study contributes to our understanding of feasibility in conjunction with the relevance and accessibility to families of CMC; however, future studies are needed to understand the feasibility and tailoring of interventions aimed to further alleviate the caregiver burden.

The care experience of the parents with the CC Team was overwhelmingly positive. The parents who were within the ITCC model of care reported to have reduced stress, disruption of daily life, and less travel time. The parents were very appreciative of the decreased burden related to care coordination and advocacy for multifaceted needs of their child, such as social and educational needs. Continuity, accessibility, and positive personalized relationships with the CC Team members were very highly regarded and valued. The parents felt heard, valued, and supported by the CC Team as partners in care of their child.

The information and communication were found to be important aspects of family experience. Improvements in the communication between the CC Team and allied health professionals were reported to have enhanced the management of CMC. Additionally, the parents indicated a strong need for proactive communication, with collaboration among all the stakeholders across different systems. This finding is consistent with other research on the vital importance of communication for parents of CMC (35) and other populations where the institutional policies regarding privacy adversely impacted the communication flow among all the stakeholders. In this respect, our pre-pandemic study illustrating the importance of accessibility *via* different communication modes is relevant to the present world of virtual care and is aligned with the call for a new "normal" in the post-pandemic care delivery (36). This new normal would include expanding the range, nature, and locations of services and supports for families as well as hybrid blended care delivery models since families still value hands-on, relationship-based, and personalized approaches.

The cost-related impact of caring for CMC is a relevant area of family experience. In the open-ended responses, the parents emphasized that extraordinary costs and lost opportunity costs cannot be captured adequately through the quantitative data. Our study underscores the extraordinary costs, which are not always medical and are associated with parenting and ensuring quality of life and optimal health outcomes for CMC. A self-directed funding and understanding what goes into the cost of raising a child with medical complexity would be important steps toward a positive change.

In addition to the intense involvement of parents in the care of their child, parents shared insights pertinent to our understanding of their engagement in research. The parents appeared to be more engaged in qualitative compared with the quantitative data collection, as demonstrated by the response rates and completion of interviews. The Family Engagement Day was very well attended and was an effective method of engaging parents in shaping the next phases of the feasibility study that include the design of interview guides. While the questionnaires had a high rate of completion, overall quantitative data collection was challenging. Recruitment rates and consent rates for the study were low, and it was difficult to engage parents to complete the follow-up questionnaires. It was evident from the qualitative data that loss to (research) follow-up was likely due to time constraints, caregiver fatigue, or limited applicability of the questionnaire items to their child. The open-ended responses of the questionnaire offered useful insights and revealed the amenability of parents to qualitative over the quantitative data collection. Furthermore, the length of the interviews (up to 90 min) points to a possible preference for more personal, narrative, and reflective forms of engagement. The mixed-methods approach was useful to explore the areas of care deemed important by parents but not captured within the questionnaire design. It also allowed for some insights into the parental engagement within different research methods. It would be useful to further integrate the design of the questionnaire used in the quantitative data collection into Family Engagement Days and one-on-one interviews. This would help to ensure that the questionnaire developed is capturing data in a way that is meaningful for parents, potentially improving the response rates. Their involvement in the questionnaire design may help to improve the response rates by making the questions more meaningful to them, and easier for them to complete.

In addition to building research capacity to capture the complex "story books" of the families (37), the balance with regard to the extent and sustainability of parent engagement should be explored in the future studies. It is known that the families of CMC are consumed with managing health of their child, which often limits their ability to engage in research (38). The multitude of often-invisible roles and tasks parents perform as caregivers, and considerations for complex daily realities of these families require further exploration. While patient and family engagement has been around for the last 20 years, the relationships of the researchers with families are still in their early stages (22). More studies are needed on the impact of patient engagement on research (39) and care, as well as guidance on engaging patients and their families (22, 40).

Several limitations of this study merit consideration. First, recruitment for the questionnaire survey was low for several reasons. Some parents were ineligible for the study due to language, social considerations, and burden of illness/stress in this population. Of eligible parents, some parents were not available for the study recruitment discussions in the clinic prior to or following the appointment of their child and others approached in the clinic did not want to take part due to already prolonged duration of complex care appointments. The transition to telephone recruitment proved effective and moderately improved the recruitment rates. Although we invited all the eligible parents on patient lists to participate in this study, the perspectives of some parents might have been missed as it is likely that those parents that are most engaged participated. Future research should aim to explore diverse methods of recruiting and engaging with parents who might not regularly participate in the research. Second, very few parents were recruited from the ITCC clinic (5 of 17 parents). A larger sample is needed from this clinic to capture a wider perspective from the parents to prevent potential bias. Third, even though the questionnaires were generally well-accepted, a few parents felt that some questions did not apply to the unique circumstances of their family, thus suggesting qualitative means may allow more nuanced ways to capture the temporal, contextual, and individual variability.

CONCLUSION

Overall, the family experience of care was generally positive for the parents of CMC. In particular, the ITCC clinic model of care offers a positive experience for the CMC and families. The ITCC clinic provides CMC and families with holistic, multidisciplinary healthcare close to their home communities, which minimizes disruptions due to travel burden and offers coordinated care between the specialists from a tertiary center and community care providers. Even though the sample size was small, it appears that the models of care may have substantial influence on the experience of care of parents. Parent engagement in research through qualitative methods allowed for richer data collection and the ability to capture information, which might have been missed through a survey. Open-ended response options in the surveys provide a means of improving survey-based engagement methods. However, we found that the participant engagement remained low despite including open-ended response options in our survey compared with the qualitative components of our study. Knowing how best to engage families of CMC in research studies is necessary for future research to understand how to evaluate and tailor the healthcare to the complex needs of families caring for CMC.

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/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Hamilton Integrated Research Ethics Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

OH analyzed the qualitative data, drafted and edited the manuscript based on the critical feedback. HS, RK, CR, and SD assisted with the data analysis and writing of the paper. PR involved at various stages in discussions about the design of the study and contributed to writing the final paper. AK assisted in the design of the study, the analysis, and critically reviewed the paper. LT provided statistical leadership, contributed to the design, and supervision of this study. AL contributed to the conception and design of the study, interpretation of the data, and revising the manuscript and edited the final version for submission. All authors contributed to the article and approved the submitted version.

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Investigating the Associations Between Child Autistic Symptoms, Socioeconomic Context, and Family Life: A Pilot Study

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Objective: The day-to-day experience of families with an Autistic child may be shaped by both, child characteristics and available resources, which often are influenced by the socioeconomic context of the family. Using a socioecological approach, this study explored the quantitative associations between child autistic symptoms, family socioeconomic status, and family life.

Methods: Data came from the Pediatric Autism Research Cohort—PARC Study (pilot). Parents of children with a recent diagnosis of autism completed a set of assessments, including the Autism Family Experience Questionnaire, Autism Impact Measure, and a Sociodemographic Questionnaire. A series of multiple, iterative linear regression models were constructed to ascertain quantitative associations between child autistic symptoms, socioeconomic context, and family life.

Results: A total of 50 children (mean age: 76 months; SD: 9.5 months; and 84% male) with data on the variables of interest were included in the analysis. The frequency of child autistic symptoms was associated with family life outcomes ($p = 0.02$ and $R^2 = 24\%$). Once autistic symptom frequency, symptom impact, and sociodemographic variables were considered, parents of higher educational attainment reported worse family life outcomes compared to their lesser-educated counterparts. This cumulative regression model had considerable explanatory capability ($p = 0.01$, $R^2 = 40\%$).

Conclusion: This study demonstrates the utility of using a socioecological approach to examine the dynamic interplay between child characteristics and family circumstances. Our findings suggest that family life for parents (of an autistic child) who have obtained higher education is reported (by the parents themselves) as less satisfactory compared to that of parents without higher education, once adjusted for the autistic symptom frequency of child, symptom impact, and income. These findings can inform the design and delivery of more family-centered care pathways during the years following a diagnosis of autism.

Keywords: autism spectrum disorder, socio-ecological framework, socio-economic context, autism symptom severity, family life

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by impairments in reciprocal social communication and social interaction, alongside patterns of restricted, repetitive behavior (1). Autism is a complex and heterogeneous condition with variability in etiology, clinical presentation, and developmental course throughout the life span (2, 3). Considering the multifactorial presentation of autism, parents and other caregivers find themselves needing to address a wide range of concerns related to child symptoms, behaviors, day-to-day functioning, and a diverse set of caregiver and family support needs (4). In some cases, behaviors such as tantrums or sleep disturbance can lead to circumstances for the family that are quite difficult to manage (4–7). The day-to-day experience of families with a child diagnosed with autism may be shaped by access to resources, such as childcare and other support systems, which often are strongly influenced by the socioeconomic context of the family (8).

To date, several studies have examined the interplay between family context (in families with Autistic children) as it relates to finances, marital relationship, and the presence of siblings. However, most of this research has focused on the quality of life of the parents and respective relatives (9–12). Although quality of life presents a useful measure to determine the state of a physical and emotional well-being of a family, these measures often do not capture the family experience holistically. We posit that a socioecological approach may be advantageous because it considers the various interrelations among individuals and their respective immediate environments (13, 14). More specifically, this conceptual framework attempts to address the dynamic interplay between the impact that the child has on the parent, and the parent on the child. Using a socioecological approach, this study explored the quantitative associations between child autistic symptoms, family socioeconomic status, and family life.

MATERIALS AND METHODS

Participants

A sample of 50 newly diagnosed preschool-aged children was recruited from April to December of 2019 as part of the Pediatric Autism Research Cohort—PARC Study (pilot) being conducted at McMaster Children's Hospital in Ontario, Canada. Participants were recruited into the pilot study *via* consecutive sampling who met the inclusion criteria of being <6 years of age (<6 years) at the time of ASD diagnosis. The data reported and analyzed in the current study were conducted when the cohort of children averaged 76 months of age. Families with insufficient knowledge of English to understand the consent process and complete questionnaires were not included in enrollment. After agreeing to be contacted about the study, consent forms were mailed out that contained a package of questionnaires. The consenting parents or legal guardians of the children with autism received a phone call to discuss the contents and instructions of the questionnaires. Families then mailed back signed consent forms with completed questionnaires in stamped business reply envelopes. This study was approved by the Hamilton Integrated Research Ethics Board (ID: 2902).

Autism Family Experience Questionnaire

The Autism Family Experience Questionnaire (AFEQ) is an ecologically valid, parent-nominated measure of family experience, quality of life, and prioritized outcomes for early intervention in pediatric autism populations (15). The AFEQ is organized into four domains: *Experience of Being a Parent of a Child with Autism*, *Family Life, Child Development Understanding and Social Relationships*, and *Child Symptoms*. For both the total score and the domain scores, a higher score is indicative of a poorer outcome. The AFEQ includes statements that are both positively and negatively worded. To best capture the association of family context, in alignment with considerations of the socioecological model, the *Family Life* subdomain was used as the primary outcome. *Family Life*, as part of the AFEQ measure, provides a quantitative value to the holistic family experience of parents, considering they have a child diagnosed with autism. More specifically, the Family Life subdomain asks parents to quantitatively document the levels of family functioning experiences that are unique to families with a child diagnosed with ASD. The Family Life domain is operationalized in the form of a Likert scale with ranges from 1 to 5 ("always" through "never") with some items being reversed scored. Examples of items in the Family Life domain include "*Family life is a battle,*" "*I feel confident to go out to family events with my child,*" "*I feel comfortable about having visitors to our home*," and "*My child is flexible in adapting to the demands of family life*." An example of a reverse-scored item is as follows: "I feel guilty about not giving other members of the family enough attention."

Autism Impact Measure

The Autism Impact Measure (AIM) is a 41-item measure that provides clinically useful information about both the frequency and the functional impact of the core symptoms of ASD (16). The measure is divided into five domains: *Repetitive Behavior*, *Communication*, *Atypical Behavior*, *Social Reciprocity*, and *Peer Interaction*. A higher score is indicative of a poorer outcome for each domain and the total score (i.e., the cumulative score of all domains). For each item, the frequency score is obtained using a five-point Likert scale ranging from 1 to 5 ("never" through "always"); the impact score is obtained by asking parents to rate the magnitude of the effect of each symptom on the everyday functioning of a child on a five-point Likert scale ranging from 1 to 5 ("not at all" through "severely"). For this study, the Frequency and Impact total scores (i.e., across all domains) were used to document the frequency and perceived impact that the core symptoms exerted on the family variables (i.e., AFEQ). To adjust appropriately for the heterogeneity of symptom presentation, the AIM was incorporated for analysis as a predictor variable for AFEQ: Family Life.

Socioeconomic Context

The family socioeconomic context considered the socioeconomic status of the parents providing care for the child with ASD. Variables of total annual income and the highest education attained were included as an adapted Hollingshead index. The total annual income in households was stratified according to the following groups: low income (0–\$39,999), medium income (\$40,000–\$89,000), and high income ($\geq$\$90,000). For educational attainment, data were organized in three levels: the "degree of high school or equivalent" was a composite outcome that included the following: no schooling, some elementary schooling, elementary schooling completed, some high school, and secondary (high) school graduation certificate or equivalent. The "degree of trade school or community college" collectively included: diploma or certificate from trade, technical, or vocational school or business college, diploma or certificate from community college, or other non-university certificate or diploma. The "degree of University or higher education" was a category comprising the following: University certificate or diploma below Bachelor's level, Bachelor's (university) degree or teacher's college, a degree in medicine, dentistry, veterinary medicine, or optometry (university certificate or diploma above bachelor level), Master's degree, and earned doctorate. The legal guardian who was responsible for filling out the questionnaire reported on their own educational attainment.

Statistical Analysis

A series of iterative multiple linear regression analyses were performed to examine the relationship between *f*amily *life* (indexed by the AFEQ subscale), child symptom severity (indexed by the AIM Frequency and Impact scale), and the socioeconomic context of the family (annual family income and highest education level attained by the legal guardian).

The following predictor variables were determined for use in the exploratory analysis *a priori*: AIM Frequency total score, AIM Impact total score, annual income, and highest degree attained by the parent of the child with ASD.

An initial model was tested in which AFEQ Family Life was regressed onto AIM Frequency. A second model was constructed to determine associations between Family Life and AIM Impact scores once adjusted for AIM Frequency scores. A final multiple linear regression model was constructed that added annual income and highest education level as variables into the model.

There were some missing data for five items (percent missing in parentheses): AFEQ Family Life (0%), AIM Frequency (3%), AIM Impact (3%), annual income (4%), and highest education level (2%). We, therefore, conducted predictive mean matching for both continuous and categorical data, creating five multiple imputed datasets using the statistical package *mice* in RStudio. Results across the five imputed datasets demonstrated little variation; therefore, the partial *F*-tests and ANOVAs were performed with the first imputed dataset. All analyses were two-tailed with a level of significance of 0.05. Statistical analysis was performed using RStudio version 1.4.1103.

RESULTS

The 50 participants in this cross-sectional study had a mean age of 76 months and a SD of 9.5 months at the time of assessment. The sample composition was 84% male ($n = 42$) and 16% female ($n = 8$). Of the 50 households sampled, a total of 47 legal guardians stated they were the mother who completed the questionnaire on behalf of the child, and three represented the father of the child. From the sample, descriptive statistics on the continuous variables of AFEQ—Family Life and AIM—Impact and Frequency, and categorical variables, namely, income and education are shown in **Table 1**. The mean of AFEQ—Family Life, AIM Impact, and AIM Frequency was 24.2, 99.36, and 122.74, respectively. The income levels (high, medium, and low) were relatively evenly distributed amongst the parents of children in the study. Regarding education, most participants had completed trades or a community college as their highest formal educational attainment at 42% ($n = 21$).

In linear regression model 1, the outcome variable of Family Life was assessed by AFEQ with AIM Frequency scores regressed as the predictor variable. Statistical associations can be found in **Table 2**. The results have indicated that AIM Frequency had a statistically significant association with Family Life (beta = 0.11, $p = 0.02$). The understanding is that for every unit increase in AIM Frequency score, the AFEQ Family score is expected to be 0.11 units greater. In addition, the model attained an adjusted R^2 of 24% with a p-value of <0.001 (see **Table 2**).

In model 2, both AIM Frequency and AIM Impact were included as predictor variables and regressed onto Family Life. Only AIM Impact displayed a statistically significant association with the outcome variable (beta = 0.07, $p = 0.04$). The understanding is that for every unit increase in AIM impact, the AFEQ Family score is expected to increase by 0.07, once AIM Frequency was adjusted for in the model. However, AIM Frequency was not statistically associated with Family Life after adjusting for AIM Impact. Overall, the model exhibited an adjusted R^2 of 29% with a p-value of <0.001. The partial F-statistic determined that once AIM Impact was considered within the nested model of model 1, the inclusion of AIM Impact exhibited a statistically significant increase in model fit (Partial F-statistic; $p = 0.0035$; see **Table 3**).

Model 3, the final cumulative model, included the predictor variables of the previous two models (AIM Impact and AIM Frequency) but was adjusted for family socioeconomic context.

TABLE 1 | Sample descriptive statistics.

Study variables	Mean	Minimum	Maximum	SD
Continuous variables				
AFEQ—family life (9–45)	24.2	10	36	5.58
AIM—impact (41–205)	99.36	41	170	29.16
AIM—frequency (41–148)	122.74	60	165	25.51
Categorical variables	***N***	**%**		
Income				
Low	14	28%		
Medium	18	36%		
High	18	36%		
Education				
High school	11	22%		
Trades/community college	21	42%		
University or higher education	18	36%		

Outcome variable	AFEQ: family life		
	Mean (SD)		**Mean (SD)**
Income		Education	
Low	22.9 (6.18)	High School	21.6 (5.89)
Medium	23.4 (4.90)	Trades/Community College	24.7 (5.83)
High	26 (5.58)	University or Higher	25.2 (4.89)

AFEQ, Autism Family Experience Questionnaire; AIM, Autism Impact Measure.

TABLE 2 | Model 1: linear regression of family life and AIM frequency.

Predictor variable	Beta coefficient	Standard error	*t*-value	*p*-value
Intercept	10.52	3.41	3.09	0.03**
AIM frequency	0.11	0.03	4.10	0.02***

*Adjusted $R^2 = 24\%$, $p = 0.0001608$***.*
$*p \leq 0.05$; $**p \leq 0.01$; $***p \leq 0.001$.

TABLE 3 | Model 2: multiple linear regression of family life, AIM frequency, and AIM impact.

Predictor variables	Beta coefficient	Standard error	*t*-value	*p*-value
Intercept	10.08	3.30	3.10	0.04**
Symptom severity				
AIM frequency	0.061	0.04	1.76	0.08
AIM impact	0.07	0.03	2.17	0.04*

*Adjusted $R^2 = 29\%$, $p =< 0.001$***.*
Partial F-Statistic ($F = 4.7135$, $df = 1$), $p = 0.035$.*
$*p \leq 0.05$; $**p \leq 0.01$; $***p \leq 0.001$.

TABLE 4 | Model 3: multiple linear regression of family life, AIM frequency, AIM impact, and socioeconomic context.

Predictor variables	Beta coefficient	Standard error	*t*-value	*p*-value
Intercept	5.73	3.34	1.72	0.093
Symptom severity				
AIM frequency	0.09	0.03	2.49	0.017*
AIM impact	0.06	0.03	2.04	0.05
Income				
Low income	Reference group	Reference group	Reference group	Reference group
Medium income	−2.25	1.68	−1.34	0.19
High income	1.08	1.63	0.66	0.51
Education				
High school	Reference group	Reference group	Reference group	Reference group
Trades/community college	1.87	1.66	1.12	0.27
University or higher education	4.48	1.71	2.62	0.01*

Adjusted $R^2 = 40\%$, $p =< 0.001$.*
Partial F-Statistic ($F = 2.9949$, $df = 4$), $p = 0.028$.*
$*p \leq 0.05$; $**p \leq 0.01$; $***p \leq 0.001$.
AFEQ, Autism Family Experience Questionnaire; AIM, Autism Impact Measure.

Once adjusted for income and education, only the predictors of AIM Frequency and parents/guardians who had pursued higher education exhibited a statistically significant association with the outcome of *Family Life*. The beta coefficient for AIM Frequency was determined to be 0.09 with a *p*-value of 0.017, whereas the coefficient for University or higher education was 4.48 with a *p*-value of 0.01. The understanding is that for every unit increase in AIM Frequency score, the AFEQ family score is expected to increase by 0.09 once AIM Impact and education are adjusted for in the model. For higher education, the understanding is that on average participants who have obtained a higher education have on average a 4.48-point increase in Family Life score (i.e., doing worse) compared to those who attained only a high school education.

The final model attained an adjusted R^2 of 40%. The remaining predictor variables were not statistically significantly associated with Family Life. The partial *F*-statistic determined that once education and income were considered in Model 2, the inclusion of socioeconomic context exhibited a statistically significant increase in model fit (Partial *F*-Statistic, $p = 0.028$), which indicates that there is sufficient association to conclude that the regression model fits the data better than the model with variables only accounting for autism symptom severity (see **Table 4**).

Evaluating the impact of socioeconomic context may present the issue of multicollinearity. For example, level of education and annual family income can be expected to exhibit some relationship to one another. The variance inflation factor (VIF) was used to examine the magnitude of multicollinearity between predictor variables. Because none of the predictor variables was found within the accepted range of 4–10, it can be assumed that no issues of multicollinearity impacted the model.

DISCUSSION

This study examined the associations between child autistic symptoms, family socioeconomic context, and family life. Using a socioecological approach and a series of multiple linear regression models, our findings demonstrate the importance of the iterative process of exploring the cumulative contribution of several factors—at both the child and family level—when trying to understand the association between symptom presence and impact, socioeconomic context, and family life. Specifically, each iteration of the regression model exhibited a considerably greater ability to explain the phenomenon under investigation. The initial model, which included only the *frequency* of core autistic symptoms, accounted for 24% of the variance explained for family life. This is an important finding which aligns with previously published research documenting similar associations between child symptom severity and parental quality of life (16–18). For example, parents of children with autism have reported experiencing a greater frequency of depressive episodes and negative emotions as the symptom severity increases (19, 20). However, it is important to acknowledge the temporal aspect of our analysis; data were collected around age 6 when the effects of symptom presentation were likely compounded with the relatively recent news of the diagnosis along with the preparation for additional and demanding responsibilities for the care of a child with autism. In the second model, where the *impact* of those symptoms was considered, 29% of the variance was explained. However, with the addition of *socioeconomic context* in the final model, adjusted for symptom frequency and impact, 40% of the variance was explained. This significant explanatory power demonstrates the utility of a socioecological approach that can offer insights into evaluating the cumulative quantitative associations among various child and family variables. Specifically, our findings highlight the importance of considering the socioeconomic context of the family above and beyond the symptom severity of a child to gain a better understanding of current family life and circumstances.

Because the evaluation of the construct of family life is closely related to other measures akin to the quality of life, it is important to assess the potential convergent validity of our findings. Studies have shown that income exhibits conflicting relationships as a predictor of quality of life and life satisfaction for parents of children diagnosed with ASD (21). In one study, the severity of ASD symptom presentation was a significant predictor of parental quality of life; however, once adjusted for family income, there was no relationship between quality of life and symptoms (22). In our current study, it was observed that once income was taken into account and adjusted for, a statistically significant relationship was observed between AIM Frequency scores and family life. By contrast, parental education level exhibited a statistically significant relationship even after adjusting for symptom frequency, symptom impact, and family income. On average, parents with higher educational attainment reported worse family life experiences than their counterparts with less education. Previous research documents effects that may provide some context for our findings. Hidalgo et al. (23) conducted an analysis of socioeconomic context on life satisfaction of parents with children with ASD and determined that mothers with a high school education or less were more likely to be satisfied with the current services and care than mothers with higher educational attainment (22, 24). The researchers speculated that parents with higher education were more aware of the diversity of autism interventions and exhibited greater concern over whether their child was receiving the optimal care. However, parents with less education were not aware of the diversity of care and thus it was speculated, for this reason, parents of higher education reported lower levels of satisfaction.

Results from the current study suggest that, compared to parents with lesser education, parents who have obtained a University degree or higher education considered their family life to be less satisfactory, once adjusted for child autistic symptom frequency, symptom, impact, and income. Previous research has suggested that raising a child with ASD can interfere with the development of a professional career of a parent, and based on that we speculate that our results may reflect parents who have attained higher education but have experienced a mismatch between their level of education and employment status and/or professional development and, as such, view their family life as less satisfying (25). An important clinical implication of our results is that service planning should be both child- and family-centered and take into account ways of achieving improved family life, such as encouraging parents to join support groups and both share their concerns and learn from experiences and perceptions of other parents.

Strengths of this study include the use of a socioecological approach, recruitment soon after initial diagnosis, a minimal amount of missing data, and lack of multicollinearity among variables under investigation, which may have affected the relatively small sample size. Two important *limitations* of this study are: (a) the small sample size for the number of predictors, which affects the power to detect differences between groups and (b) the cross-sectional nature of the data. An additional limitation involves the timing of data accumulation analysis. A recent diagnosis of autism for parents is a difficult and often burdening moment in their lives. Considering that the data were collected near the time of diagnosis, these findings may not generalize to parents who have begun to develop effective routines and coping strategies. Large, longitudinal mixed-method studies containing follow-up assessments of both quantitative and qualitative components are required to further explore the study findings. Conducting interviews with parents of children with ASD would better provide researchers the opportunity to understand the variability in family life between households rather than relying solely on quantitative data from a set of static questionnaires.

Using a socioecological approach and a series of multiple linear regression models, this study explained considerable variance in family life after accounting for the child's autistic symptoms and family socioeconomic context. Our results suggest that family life for parents (of an autistic child) who have obtained a University degree or higher education is considered (by parents themselves) less satisfactory compared to that of parents with less education, once adjusted for child's autistic symptom frequency, symptom impact, and income. The significant explanatory power demonstrates the utility of a socioecological approach that can offer insights into the cumulative quantitative associations among various child and family variables and family life that has clinical implications.

In line with the evidence-based care model outlined by Sackett et al., our study highlights the importance of considering family socioeconomic context when planning clinical care and allocation of service resources (26). Such an approach would inform the design and delivery of more family-centered, holistic yet pragmatic, and feasible care pathways that consider both, the characteristics of the child and the family circumstances during the years following a diagnosis of autism.

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 human participants were reviewed and approved by McMaster Research Ethics Board McMaster University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

All authors have contributed significantly to the content of the article and have read and approve of the manuscript submission.

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REVIEW
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Child Maltreatment and the Child Welfare System as Environmental Factors in the International Classification of Functioning

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The WHO defines child maltreatment as any form of neglect, exploitation, and physical, emotional, or sexual abuse, committed against children under the age of 18. Youth involved in the child welfare system report more maltreatment experiences and environmental turbulence (e.g., number of moves, caseworkers), placing them at greater risk for poorer physical and mental health. The International Classification of Functioning, Disability, and Health (ICF) provides a framework to describe health conditions and severity of disabilities for an individual and/or group in the context of environmental factors. The Maltreatment and Adolescent Pathways (MAP) study is a longitudinal study, assessing self-reports on variables (e.g., child maltreatment history, trauma symptoms, dating violence, and substance use) of youth in an urban child protection service system. This study focuses on 11 of the 24 MAP publications that pertain to health and functioning, which can be considered applicable to the ICF framework, following established linking rules. The purpose of this study is to analyze these MAP sub-studies, with maltreatment and involvement in the child welfare system as environmental factors that impact the functioning of child welfare-involved youth. Findings indicate significant relationships across environmental factors (i.e., child maltreatment histories, child welfare system involvement), health conditions (i.e., trauma symptomatology, psychological distress, intellectual disabilities), and functioning problems (i.e., substance use, adolescent dating violence, sexual risk-taking, coping motives, sleep problems). The interrelated nature of these factors in the MAP sub-studies suggests the value of the ICF model to a holistic health view of use to practitioners supporting system-involved youth, clarifying unattended environmental factors in guiding service provision for foster care and/or maltreated youth.

Keywords: child welfare, foster care, child maltreatment, mental health, functioning, youth, adolescence, disability

INTRODUCTION

Child maltreatment is a significant public health issue. While most countries have child welfare systems, many maltreated youths are undetected by practitioners, and practitioners experience hesitation in fulfilling mandatory reporting duties [e.g., (1)]. According to the WHO, child maltreatment is defined as: "All forms of physical and/or emotional ill-treatment, sexual abuse, neglect or negligent treatment or commercial or other exploitation, resulting in actual or potential harm to the child's health, survival, development, or dignity in the context of a relationship of responsibility, trust or power" [(2); refer to **Table 1** for definitions of types]. In North America, the median prevalence of physical abuse was 24.3% for boys and 21.7% for girls. The prevalence of sexual abuse was 14.1% for boys and 20.4% for girls. Emotional abuse was 28.4% for boys and 23.8% for girls. Median rates of neglect differed substantially between girls (40.5%) and boys (16.6%) (4). It is well-recognised that poly-victimisation describes many youths in the child welfare system and is related to poorer outcomes (5). Studies have demonstrated a dose-response relationship, where an increased number of childhood maltreatment experiences correlated with elevated dysfunction (6, 7). While estimates of the cost to the victims and their communities are unquantifiable, the service system costs have been estimated. Based on official reporting data, the cost of non-fatal child maltreatment in 2015 was estimated to be $830,928 per case, and fatal outcomes cost $16.6 million in the United States (8). Furthermore, the total economic burden in the United States for substantiated incident cases was $428 billion in 2015 (8). It is clearly established that maltreatment yields lifespan impact to physical, mental, and financial health.

The victim of child maltreatment is probabilistically more likely to face challenges, with greater risk of poverty, and the likelihood of finding themselves homeless, couch-surfing within their communities, or street-involved (9). Living in such circumstances brings another level of harm risk, such as increased access to a greater array of substances, opportunities to make money by selling substances, increased likelihood of violence victimisation, as well as vulnerability to recruitment for sex trafficking (10). Child maltreatment is associated with multiple negative outcomes, most notably in the areas of mood disorders (e.g., anxiety, depression), substance use problems, relationship violence, and chronic physical health conditions or non-communicable diseases, as stress pathways are triggered with cascading potentials (11–16). In addition to adverse events being more prominent, there is also a lack of positive or benevolent childhood events (e.g., presence of a positive person in which to confide, opportunities for school achievement, secure parent-child attachment relationships) (17). There has been much discussion over poverty risk and child neglect, as families with socioeconomic disadvantage come to the attention of the child welfare system to a greater extent than more affluent families (18). If child welfare system engagement creates a safety net around the child, mandatory reporting may be considered part of prevention planning for re-victimisation risk (1). In over 30 countries, mandatory reporting laws direct suspected abuse and neglect events or risks thereof to the responsible child welfare authorities (19). The most comprehensive laws and policies are in the United States, Canada, and Australia, yet many other high-income countries do not have such laws as policy (e.g., Hong Kong, Germany, Hungary, etc.). Entry into the child welfare system may bring a wide range of financial, medical, and other social services to the child and/or family. As a foster care youth living in an out-of-home arrangement, there may be a linkage to a caring home and a stable caseworker, or there may be experiences of revictimization in care and a prolonged lack of permanency (20), multiple residential transitions (21, 22), as well as multiple changes in caseworkers (23). According to Waid et al. 40.8% of youth in foster care had experienced at least one placement change throughout 18 months (24). Furthermore, caseworker turnover rates have been reported to be 30–40% annually (25). However, the system-specific risk may be heightened when a youth "ages out of care" in adolescence or early adulthood, potentially without transition services and a social safety net.

TABLE 1 | Definitions of childhood maltreatment types (3).

Term	Definition
Physical abuse	"A caregiver inflicting physical harm or engaging in actions that create a high risk of harm"
Emotional abuse	"Inflicting emotional harm through the use of words or actions"
Sexual abuse	"Any action with a child that is done for the sexual gratification of an adult or significantly older child"
Neglect	"Failure to meet a child's basic physical, emotional, educational, and medical needs"

Maltreatment, Adolescence, and Child Welfare

According to the United States Department of Health and Human Services (USDHHS) Child Maltreatment 2019 report (26), infants (up to age 2) are the largest group for entry into foster care, representing 28% of all cases, or a rate of 26.7 per 1,000 children (27). Youths aged 12 and older are the next highest timeframe for coming into care (28). As of 2020, the number of youths in care in the United States is ∼424,000 (29). The WHO defines adolescence from 10 to 19 years of age, which is noted as a critical development period. With the onset of puberty, increases in sex hormones contribute to rapid changes in physical growth, the brain, and behaviour (30). In terms of maltreatment types, adolescence is a higher time of sexual violation among females: ages 14 and above represented about 72% of sex trafficking cases in child welfare (27). Thus, it is important to focus on child sexual abuse (CSA) when considering the functioning of adolescents, in terms of risk, as well as in terms of its potential impact on the adolescent developmental task of becoming attuned to and involved in romantic relationships. Furthermore, adolescence is a known high period of cognitive development, wherein risk-taking, and abstract and strategic thinking becomes refined (31). Studies have highlighted the potential negative impact of stressful life experiences during adolescence, which may disrupt normal neural development and contribute to adolescent vulnerability to mental diseases, such as anxiety and/or depression (32–34). Among children investigated by child protection services (CPS) in the National Survey of Child and Adolescent Well-being (NSCAW) II, approximately half were noted as having developmental issues (35). As compared to children without disabilities, child welfare-involved children with intellectual disabilities were at greater risk of experiencing placement instability, challenges with adoption, and not being able to reunite with kin (36). Child welfare-involved youth are also at higher risk for re-victimisation in their dating relationships (37). Dating violence victimisation is associated with self-reported post-traumatic stress symptomatology (PTSS), drug use, and previous dating violence perpetration (37–39).

While entry into child welfare indicates a sentinel maltreatment risk event, there is a wide range of experiences that may occasion re-victimisation, such as visitation with kin or parents, experiences in the alternate care environment, street involvement, and the use of restraints in institutional care. This potential for further trauma needs to be regarded as an active "rule out" when child welfare youth are cared for in the medical home (40). As youth-reported maltreatment experiences have been shown to be higher than official recordings of victimisation, it is critical to ask youth directly about their victimisation experiences [e.g., (41)].

The International Classification of Functioning, Disability, and Health

The International Classification of Functioning, Disability, and Health (ICF) by the WHO provides a standardised language and framework for describing an individual's health, functioning, and disability (42). The guiding document for definitions, categories, and codes for the ICF is the ICF Red Book (42). The ICF is a hierarchal classification, based on the biopsychosocial model which views functioning as an outcome of the dynamic interactions of both health conditions (e.g., diseases, disorders, and injuries) and contextual factors (e.g., social, child welfare system involvement) (42). The ICF is structured into two parts: (1) functioning and disability, and (2) contextual factors. Within these two parts are six levels ordered as a hierarchy, from general to more detailed and specific entities: components, domains, constructs, positive aspects, and negative aspects. Functioning and disability include these components: body functions (e.g., sleep, pain, and emotional functions) and structures (e.g., nervous, reproductive, and endocrine system), as well as activities (e.g., basic learning, self-care, and mobility) and participation (e.g., communication, interpersonal relationships, and education). Contextual factors include the components: personal (e.g., sex, age, and ethnicity) and environmental (e.g., technology, social support, and policies) factors (43). Domains, constructs, positive aspects, and negative aspects progressively detail the aspects of functioning. This study will focus on the components of functioning and disability and contextual factors.

As applied to an individual, the ICF can provide a snapshot in time that allows one to consider a wide array of variables to obtain a more holistic analysis of the individual's historical factors and current functioning. This model may be particularly beneficial in the context of child maltreatment. For example, youth with higher exposure to maltreatment report higher levels of chronic pain (44). Pain in identified locales, or chronic generalised pain, would appear in the ICF body functions domain. When encountering youth in chronic pain without a clear medical condition, the differential diagnosis may be to rule out child maltreatment history, noting most victims do not disclose (45). The ICF model would be more likely to detect this child maltreatment history as a barrier to functioning, due to its more holistic approach. The ICF is, therefore, well-suited to frame the "whole" youth in terms of inter-connecting domains of functioning, for example, multiple problems that may ensue from the presence of post-traumatic stress symptoms (PTSS) of arousal (e.g., sleep disturbance, sexual risk-taking/multiple partners), negative cognitions and mood (e.g., depression), avoidance (e.g., substance abuse) and re-experiencing (e.g., re-victimisation) (46). The principle of multi-finality (47) reflects that many outcomes may arise from a common event (e.g., maltreatment experiences as the abuse of power and negligence).

The application of the ICF to conceptualising or matching service needs within child welfare has been under-utilised. Most of the research in child welfare has focused on rehabilitation services, related to physical or cognitive disability, to the relative exclusion of mental health and related problems as disability or risk thereof. Only one child welfare study was found, to our knowledge, that explored disability (i.e., physical, medical, intellectual), as well as mental health conditions *via* the ICF framework (48). Indeed, the overlap with a physical and cognitive disability, and other areas of functioning compromise, is an area of research gap. It is critical to take into account disability in terms of complex trauma or ongoing mental health challenges, which facilitate other problems such as alcohol use and dating violence. As such, a focus on adolescent functioning within child welfare represents a valid start to considering the application and utility of an ICF approach, for integration across disability types, mental health problems, and environmental factors. It must also be considered that many child welfare system-involved youth show remarkable resilience (49). The progression through child welfare may involve case openings and closings before an out-of-home care option is sought permanently. Thus, the ICF application may be best understood in terms of distal or historical domains, compared to current or proximal influences.

In this study, we review a selection of studies that use data from the first comprehensive research study on child welfare system-involved youth in Canada, known as the Maltreatment and Adolescent Pathways (MAP) study. This longitudinal study captures data from adolescents in a large urban CPS system in Ontario, Canada, with study entry between ages 14 and 17, and followed up to 3 years. Most youth will exit the child welfare system in the age ranges of 16–18 years, with some extending support to age 21. By investigating these MAP sub-studies in the context of the ICF framework, we aim to explore the complex interactions of factors that affect the functioning and health of maltreated, system-detected youth and propose a model of distal and proximal considerations. Given the mobility of child welfare-involved youth and the rapidity with which most cases are closed or transferred to other services, a functioning-oriented approach seems essential to guide youths' health and well-being, wherever they are at in their trajectory through child welfare services. The goal herein was to examine the MAP set of findings and identify those that could be considered within the ICF framework to illustrate maltreatment and the child welfare system involvement as environmental factors, in conjunction with mental health problems and disabilities as health conditions. While the MAP study did consider selected resilience factors [e.g., self-compassion (50)], and these should be integrated into an ICF framework, they were not the focus of the current exploration.

METHODS

The Maltreatment and Adolescent Pathways (MAP) Study

A multi-disciplinary research team, guided by a child welfare advisory board, developed and implemented the MAP study. The MAP study collected information about physical health (e.g., sleep quality), mental health (e.g., PTSS), and cognition (e.g., IQ and memory) in adolescents from a large urban CPS system catchment area. The MAP study recruited participants drawn *via* a random numbers table from a CPS agency-provided list of all active caseloads of adolescents (average age = 15.8 years), refreshed every 6 months given the rate of case closures. These included the largest agencies in Canada (50, 51).

Among the 24 MAP sub-studies, 11 were selected for analysis due to their fit with the ICF framework. The fit was determined based on whether the sub-study investigated data on health conditions, contextual factors, and functioning or disability of child welfare-involved youth, so it could be holistically applied to the ICF framework. A description of each included MAP sub-study can be found in **Table 3**. Linking rules were used to connect the concepts in MAP sub-studies with the appropriate ICF categories and to enhance the transparency of the linking process (52, 53). A paediatric consultant, with expertise in the ICF, supported our linking procedures. The MAP sub-studies use a diverse range of measures, as seen in **Table 2**, and our goal is to link these measure outcomes to ICF categories, with the focus on childhood trauma exposures and adolescent mental health and resilience. The linkage to the ICF was completed by an ICF specialist, child welfare/mental health specialist, and medical student with a background in the study of maltreated youth.

Health conditions in the ICF refer to any diseases, disorders, and/or injuries, which may include other circumstances such as stress, ageing, pregnancy, congenital anomality, and genetic predisposition (42, 43). In this study, we identified mental health problems (trauma symptoms and psychological distress) and intellectual disabilities, as measured in the MAP study, such as health conditions. Contextual factors in the ICF include environmental and personal factors, which facilitate or act as barriers to functioning. For the purposes of this study, environmental factors included: child welfare system experience (length of time in the system, foster care or non-foster care, number of residential moves; e5258), caseworker relationship, (number of visits, identification with caseworkers; e360), child maltreatment experience (e310, e398), and exposure to intimate partner violence (IPV; e310, e398) as measured by the Childhood Experiences of Violence Questionnaire [CEVQ; (57)]. Personal factors included sex, age, ethnicity, attachment style, and past experiences (youth-reported history of child maltreatment types) and were coded as personal factors (pf). Activity limitations represent the difficulties an individual would have in executing any given task or action (e.g., walking, eating). Participation restrictions encompass the problems an individual may experience with others during the involvement of life situations (e.g., employment, education). Activity limitations and participation restrictions are considered one component due to their correlated nature. Among youth in child welfare, we have identified substance use (d5702), sexual risk-taking (d5702), and adolescent dating violence (ADV; d7202, d7700) in this component. While ADV may fall under contextual factors (i.e., current victimisation can be environmental; victimization; and/or perpetration history can be personal), they are defined as participation restrictions for the purposes of this study in so far as they reflect the abuse of power and control, with potential limits to the access of other peers and family (socially isolating the partner), and capacity to exit a romantic partnership and/or begin new ones (69). Romantic relationships are an important and under-considered domain of the child welfare system-involved youths' functioning. Especially given the relative lack of close relationships and supportive family for child welfare-involved youth, there may be an early entry into these relationships with peers or older adults (70). Finally, body functions describe the physiological and psychological functions of body systems. We have placed coping motives (b1301) and sleep disturbances (b1340, b1341, b1342) in this component. Coping motives were specifically considered mental functions, as they encompass the motivations to engage in certain behaviours. Body structures (i.e., anatomical body parts) were not explored in this review.

The ICF offers a snapshot in time of functioning. Youth in the child welfare system context, however, face instability, such as frequent moves, varying lengths of placements, and recurring victimisation at different time points. Such instability contributes to the need for an analysis of proximal (now or recent) and distal (historical or lifetime) factors that impact the functioning of child welfare-involved youth. For the purposes of this study, proximal factors include factors measured currently or within the past year. Distal factors include factors measured beyond a year or based on frequency, therefore being lifetime (Refer to **Figures 1**, **2** for our distribution of factors from the MAP study, respectively).

It is important to note that the listed factors measured by the MAP study are not exhaustive of all relevant variables. For example, sexual orientation was reported by youth and fluctuated across MAP assessments over time, which is consistent with the concept of sexual identity exploration, with an overall higher representation of sexual minority status than expected in the population (75, 76). Sex at birth, gender identification, and sexual orientation are currently considered in more expansive categories than were evaluated in the MAP study [e.g., transgender, non-binary, gender queer, etc. (77)]. While sex

TABLE 2 | MAP study measures.

Outcome	Measure(s) used	Corresponding code
Child maltreatment (i.e., physical abuse, sexual abuse, emotional abuse, neglect)	Childhood Trauma Questionnaire [CTQ; (54, 55)] • 70-item self-administered inventory with 5-point Likert-type scales • Addresses five types of maltreatment (emotional abuse, physical abuse, sexual abuse, emotional neglect, and physical neglect) CTQ-short form [CTQ-SF; (56)] • 28-item version of the original CTQ	e310 (immediate family), e398 (support and relationships)
Exposure to intimate partner violence (IPV) (i.e., physical and verbal)	Childhood Experiences of Victimisation Questionnaire [CEVQ; (57)] • Brief 18-item self-report measure of victimisation among adolescents • Addresses peer-on-peer violence, witnessing domestic violence, emotional abuse, physical punishment, physical abuse and sexual abuse	e310 (immediate family), e398 (support and relationships)
Adolescent dating violence (ADV)	Conflict in Adolescent Dating Relationships Inventory [CADRI; (58)] • 46-item self-report questionnaire, with bidirectional questions (victim/perpetrator) • Measures abusive behaviour among adolescent dating partners	d7202 (regulating behaviours within interactions), d7700 (romantic relationships)
Trauma symptoms (i.e., anger, anxiety, depression, dissociation, sexual concerns, PTSS)	Trauma Symptom Checklist for Children [TSCC; (59)] • 54-item self-report of trauma symptoms • Assesses severity of post-traumatic stress and related psychological symptomatology (anxiety, depression, dissociation, anger, sexual concerns)	Health conditions are coded by ICD-10 and not ICF
Psychological distress	Brief Symptom Inventory [BSI; (60)] • 53-item self-report of clinically relevant psychological symptoms in adolescents and adults • Addresses somatization, obsession-compulsion, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, and psychoticism	Health conditions are coded by ICD-10 and not ICF
Substance abuse (i.e., alcohol misuse, marijuana misuse, use of drugs)	Rutgers Alcohol Problem Index [RAPI; (61)] • 23-item self-administered screening tool for assessing adolescent problem drinking Alcohol Use Disorders Identification Test (AUDIT; (62)) • 10-item questionnaire covering the domains of alcohol consumption, drinking behaviour, and alcohol-related problems CRAFFT (63, 64) • Substance use screening tool for adolescents	d5702 (maintaining one's health)
Sleep disturbances (i.e., taking longer than half an hour to fall asleep, waking up before intended, having non-restorative sleep)	11 self-report questions adapted from standardised sleep disorders questionnaire [SDQ; (65)] SDQ • Short self-rating questionnaire with 18 questions on different sleep problems	b1340 (amount of sleep, b1341 (onset of sleep), b1342 (maintenance of sleep)
Intelligence [i.e., intelligence quotient (IQ)]	Kaufman Brief Intelligence Test [KBIT; (66)] • Individually administered measure of intelligence • Developed specifically for screening and related purposes	Health conditions are coded by ICD-10 and not ICF
Drinking motives	Drinking Motives Questionnaire Revised [DMQ-R; (67)] • Self-reported questionnaire with 20 reasons why people might be motivated to drink alcoholic beverages • Addresses social, coping, enhancement, and conformity motives	b1301 (motivation)
Sexual risk-taking	US Youth Risk Behaviour Survey [YRBS; (68)] • Monitors health-related behaviours among youth and adults (e.g., alcohol use, unhealthy dietary behaviours, inadequate physical activity)	d5702 (maintaining one's health), b1301 (motivation)

may be a distal category, gender may be better considered as a proximal variable.

Among the included 11 sub-studies, data were extracted independently by three authors (KK, CM, and JP). For each study eligible for full-text assessment, the number of participants, characteristics of participants (e.g., age, sex, etc.), health conditions, and components of the ICF framework (i.e., environmental factors, personal factors, body functions, activities, and participation) were identified. Any discrepancies regarding these extractions were resolved by discussion among the authors.

RESULTS

A total of 11 publications were included in our selective review. All 11 publications were published between 2009 and 2019, and used the same MAP study dataset, but varied in their selection of data to analyze (available complete data). Sample sizes ranged from a select sample of 73 (i.e., a sample of youth who completed IQ testing) to its full sample of over 500 subjects. Borderline to mild intellectual disabilities (defined as IQ scores between 60 and 84) were present in 24% of the Weiss et al. (78) study. All analyses reflected more females than males (ranging from 51.8 to 64.4%

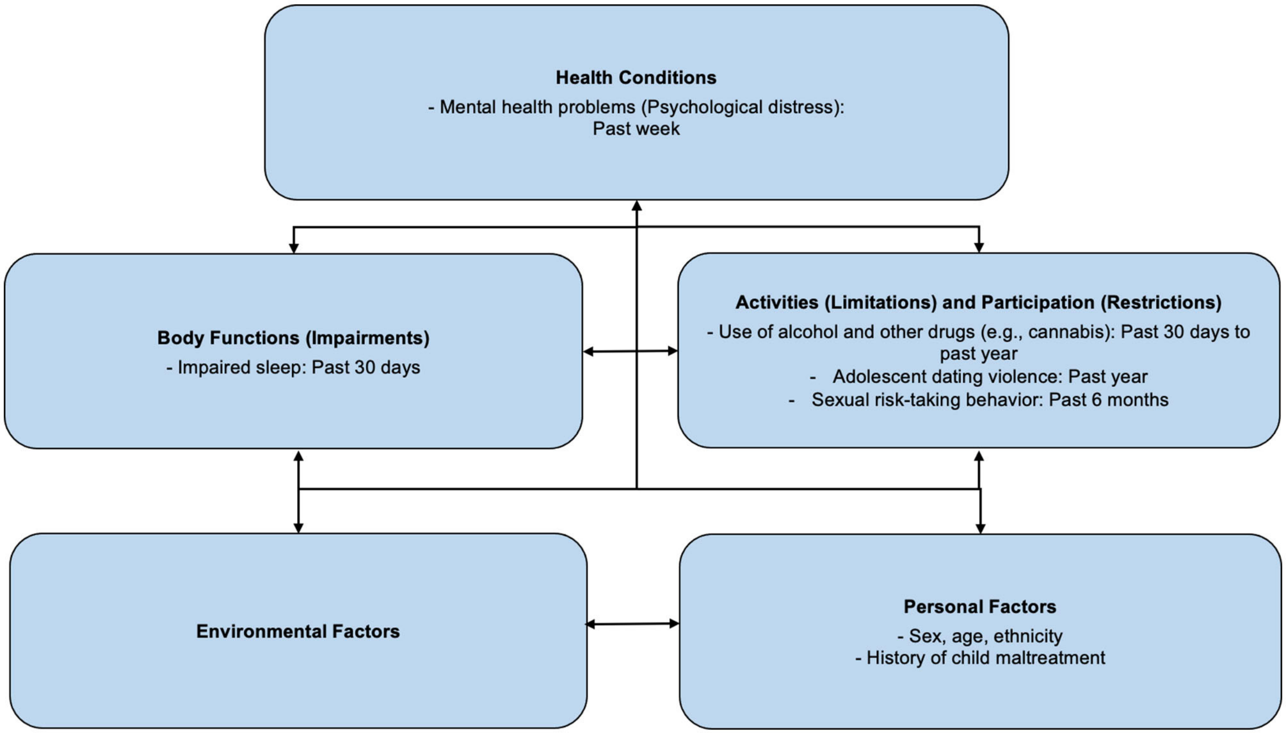

FIGURE 1 | Distal ICF factors: Sexual risk-taking behaviours include early sexual debut, multiple sexual partners historically, a pattern of unprotected intercourse, or sex while under the influence of alcohol or drugs. These may result in negative personal health implications (71). Impaired sleep refers to the presence of difficulty with sleep, which could present itself as sleep onset latency, frequent nocturnal awakenings, or prolonged periods of wakefulness during the sleep period (72).

of participants). Refer to **Table 3** for included sub-studies and additional study characteristics descriptions.

Contextual Factors

The ICF model postulates a dynamic interaction across health conditions, contextual factors, and functioning. The influence of personal and environmental factors presents across the different domains of functioning and disability. Our focus is on child maltreatment as an environmental factor.

Maltreatment Experiences

Maltreatment and Adolescent Pathways (MAP) youth reported high rates of maltreatment distally. In McPhie et al. (51), 78.1% of the participants reported one or more types of maltreatment histories above the Childhood Trauma Questionnaire [CTQ; (56)] clinical cut-offs for severity. Rates and severity of child maltreatment histories did not differ based on participant IQ, or Indigenous identity (78, 84). In Hudson et al. (81), 37% of participants self-reported CSA histories. CSA histories emerged as a particularly impactful form of maltreatment: it was related to the severity of all types of child maltreatment for both sexes (76). Faulkner et al. (75) investigated both maltreatment histories and lifetime exposure to IPV. In this analysis, 64.6% of participants reported two or more types of childhood maltreatment histories. Respectively, 60.8 and 38.6% of individuals witnessed lifetime verbal violence and physical IPV, according to the CEVQ. High levels of poly-victimisation rates of maltreatment were confirmed in this child welfare-involved sample.

Child Welfare System Experiences

Most MAP youths had received long-term services, in the 5–6-year range, with mid-childhood (ages 9–12), as the most common entry time into care. Multiple placements were the norm: youth in the MAP study moved in the range of 0–5 times in the past 5 years (averaging at 2.1 times). The provision or level of treatment or referral was not known; in this Ontario sample, there were mandated caseworker visits every 90 days to the youth. However, caseworker compliance and the length and nature of caseworker visits were not known. Only identification with caseworkers was investigated in Waechter et al. (84), and it did not differ between Indigenous and non-Indigenous child welfare-involved youth. In this study, the question of whether positive identification (as compared to negative identification) with a caseworker was considered to understand to some degree the nature of the relationship.

Health Conditions

Mental Health Problems

In terms of the relationship of maltreatment types to health conditions, there was a range of significant findings across mental health conditions, with child emotional and sexual abuse showing higher levels of adolescent concern. McPhie et al.

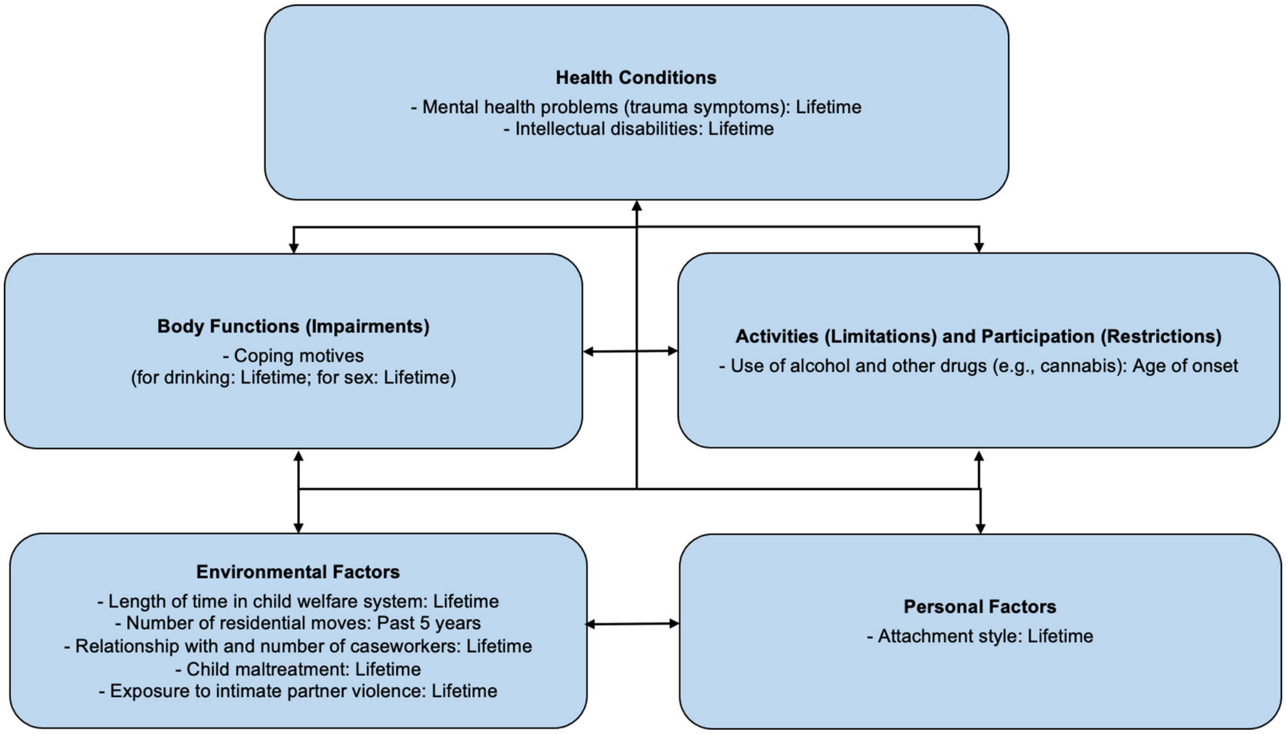

FIGURE 2 | Proximal ICF factors: Coping motives refer to the process governing the choices made towards various coping behaviours, in an effort to manage stress (73). Maladaptive coping motives may lead to health-risk behaviours (e.g., drinking alcohol to cope with negative affect) (74).

(51) found that 28.2% of the sample reported one or more symptoms of psychological distress within the clinical range in the past week on the BSI measure, which is a proximal distress measure [domains of somatization, anxiety, depression (60)]. Goldstein et al. (80) and Park et al. (82) reported that all types of child maltreatment histories (i.e., physical abuse, sexual abuse, emotional abuse, physical neglect, emotional neglect) had positive, significant correlations with the trauma symptoms, in terms of the TSCC (59) total score, as well as the subscales of PTSS, dissociation, and sexual concerns. As the TSCC does not specify a timeframe, it may be conservatively regarded as reflecting lifetime, rather than current trauma symptoms. Emotional abuse history was a strong predictor of TSCC total score in Wekerle et al. (85), when all other forms of historical maltreatment were taken into account statistically, indicating a unique contribution. CSA history was associated with increased anxiety, depression, anger, and PTSS (81). Trauma symptoms did not differ based on Indigenous identity (84).

Activity Limitations and Participation Restrictions

Adolescent Dating Violence (ADV)

Adolescent dating violence (ADV) was assessed in the timeframe of a romantic partnership in the past year, and a dating relationship was defined as extending 2 or more weeks, based on relationship prevalence information (58). Over 60% of the MAP sample (aged 14–17 years old) endorsed having begun or been in a dating relationship. While the majority of child welfare-involved youth reported being in a relationship, many were also not reporting a relationship status. Of a 158-participant sample, 77.2% reported perpetrating verbal/emotional ADV, 36.7% threatening violence, 32.9% using physical violence, and 11.4% using sexual violence (75). In Wekerle et al. (85), among MAP dating youths, the majority experienced either form of ADV (i.e., over 60% of females; over 40% of males), noting that perpetration and victimisation behaviours total scores were positively correlated. Male participants with non-foster care status had higher ADV victimisation, as compared to foster care male youth, whereas ADV perpetration did not differ between sexes or foster care status (83). ADV over the MAP assessments were considered in Tanaka et al. (83). From a longitudinal perspective, the minority of child welfare-involved youth (33.6%) were never exposed to ADV, from initial data collection to 2-year follow-up. About 46% of youth reported experiencing repeated ADV (2+ times across assessments), although whether the romantic partner remained the same or varied could not be determined. In terms of types of ADV, verbal and/or emotional abuse was most highly endorsed, and sexual abuse was least endorsed. This is consistent with the view of ADV as a potential participation restriction.

Child maltreatment histories, trauma symptoms (i.e., anger and anxiety), intellectual disabilities, and foster care status were associated with both victimisation and perpetration of ADV. In Wekerle et al. (85), considering sex differences, trauma symptoms

TABLE 3 | Study characteristics.

References	Aim of study	Sample	Gender (% female)	Mean age (years) [SD]	Main findings
Faulkner et al. (75)	To examine the relationship between experiencing two child maltreatment types (child abuse and neglect [CAN] and exposure to IPV) and two outcomes (substance use and dating violence) in the past year.	$N = 158$ Participants remaining in the MAP study at the two-year follow up and who had initiated dating.	62.7%	17.89 [0.98]	- CAN experiences predicted more frequent dating violence perpetration and greater alcohol problems, indirectly via anger. - Exposure to caregiver IPV was associated with dating violence, indirectly via anxiety and anger. - Exposure to caregiver IPV was associated with marijuana use, indirectly via anxiety and dissociation. - Exposure to caregiver IPV was associated with past year occurrence of both marijuana use and dating violence, indirectly via anger
Goldstein et al. (79)	To develop a framework that identifies risk of alcohol problems and the likelihood of benefiting from preventive interventions among child welfare-involved adolescents.	$N = 202$ Participants who had consumed alcohol in the past year and had complete data on the variables of interest.	54.5%	15.93 [1.03]	- Child maltreatment was significantly and positively associated with alcohol problems and with coping motives. - Coping motives were significantly and positively associated with anxiety symptoms and with alcohol problems. - Increased anxiety symptoms were associated with more alcohol problems for adolescents with high coping motives compared to low. - Increased depression symptoms were associated with fewer alcohol problems among those with high coping motives compared to low.
Goldstein et al. (80)	To examine the contribution of post-traumatic stress symptoms to substance use and substance related problems among child welfare-involved adolescents and emerging adults.	$N = 253$ Participants who had complete data for variables of interest at the initial, 6-month and 1-year assessment points.	61.4%	16.87 [1.04]	- All maltreatment types were positively and significantly associated with all trauma symptoms in TSCC measure (except for the association between sexual abuse and anger). - Child maltreatment and dissociation were positively associated with using a greater number of illicit drugs (other than marijuana) in the past year. - Anger and dissociation were positively associated with alcohol use and drug problems. - Sexual concerns were negatively associated with drug problems.
Hudson et al. (81)	To examine the gender differences within the links between CSA and alcohol problems in adolescence, via potential emotion-focused mechanisms, among child welfare-involved youth.	$N = 301$ Participants who had complete data on CSA, negative emotion symptoms, and problem drinking questionnaire items.	56%	16.4 [1.0]	- CSA was positively associated with increased levels of anxiety, depression, and anger. - CSA was indirectly associated with problem drinking, via anxiety and anger among female adolescents (full mediation by negative emotions). - CSA was indirectly associated with problem drinking, via anger (partial mediation by negative emotions).
McPhie et al. (51)	To examine the relationship between child maltreatment history and sleep quality among adolescents.	$N = 73$ Participants in the initial and 2 year time points with complete data on variables of interest.	64.4%	15.9 [1.06]	- Initial severity of child maltreatment predicted sleep problems 2 years later. - Psychological distress fully and positively mediated the relationship between child maltreatment and sleep problems.

(Continued)

TABLE 3 | Continued

References	Aim of study	Sample	Gender (% female)	Mean age (years) [SD]	Main findings
Park et al. (82)	To examine how PTSS and coping motives mediate the association between child maltreatment and alcohol use.	*N* = 564 Participants with complete data on childhood trauma, PTSS, drinking motives, and alcohol misuse at initial data collection and 6-month follow-up.	53.7%	15.9 [1.1]	- Child maltreatment was positively correlated with PTSS. - PTSS were positively correlated with coping motives. - Coping motives were positively correlated with alcohol misuse. - PTSS and coping motives mediated the relationship between child maltreatment and alcohol misuse via a serial mediation (i.e., not as single mediators).
Tanaka & Wekerle (83)	To examine self-reports of ADV victimisation and perpetration among child welfare-involved youth.	*N* = 341; 110 Participants who completed the ADV measurement at initial data collection; participants who completed the ADV measurement across all data points.	54%	15.8 [1.1]	- ADV verbal/emotional abuse was most highly endorsed. - ADV sexual abuse was least endorsed. - No significant difference in ADV perpetration and victimisation scores and prevalence across genders. - Male ADV victimisation was higher for non-foster care youth compared with foster care youth; male ADV perpetration did not differ. - Females did not differ based on CPS status. - 33.6% of youth have never been exposed to ADV from initial data collection to the 2 year follow up. - 46.4% reported ADV 2+ assessment points, up to the 2 year follow up.
Waechter et al. (84)	To examine the relationship between cannabis use and self-reported identification with a caseworker among Indigenous and non-Indigenous adolescents.	*N* = 476 Participants who self-identified as Indigenous or other.	53%	15.8 [0.99]	- Indigenous youth did not differ from the non-Indigenous youth on their child maltreatment types, IPV, PTSS, cannabis usage, nor identification with CPS workers. - Indigenous youth who reported low identification with their caseworker were 5.47 times more likely to have ever used cannabis in the past 12 months compared to non-Indigenous youth with low identification.
Weiss et al. (78)	To examine attachment styles and ADV in child welfare-involved adolescents with borderline-to-mild intellectual disability.	*N* = 167 Participants who completed intelligence testing and experienced clinically significant maltreatment histories.	58%	15.8 [0.98]	- Adolescents with borderline-to-mild intellectual disabilities reported significantly more ADV victimisation and perpetration than adolescents with average IQ. - Rates and severity of child maltreatment histories were similar across adolescents with intellectual disabilities and with average IQ. - Avoidant attachment style significantly predicted ADV victimisation and perpetration, particularly among adolescents with lower IQ.
Wekerle et al. (76)	To examine sexual motives, CSA, and risky sexual behaviour among child welfare-involved youth. To also evaluate motivations for sexual behaviour as a potential mechanism from CSA to risky sexual behaviour among adolescents.	*N* = 297 Participants who endorsed being sexually active (defined as having had sexual intercourse at the time of initial assessment).	57.6%	15.83 [1.04]	- CSA was associated with severity of all child maltreatment types for both genders. - CSA was associated with witnessing emotional IPV for females only. - CSA was associated with more sexual risk taking, particularly among males, compared to youth with no CSA experiences. - CSA was associated with greater coping motives, which in turn was associated with increased sexual risk-taking.
Wekerle et al. (85)	To consider the predictive value of childhood emotional abuse to understand PTSS and ADV. To also assess PTSS as a mediator between childhood emotional abuse and ADV.	*N* = 402 Participants who had data on childhood maltreatment histories, PTSS and ADV at initial data collection of the MAP study.	51.8%	16.3 [0.99]	- Emotional abuse significantly predicted both PTSS and dating violence among males and females. - PTSS significantly mediated the relationship between male emotional abuse and ADV perpetration. - PTSS significantly mediated the relationship between female emotional abuse or physical abuse and ADV victimisation.

mediated the relationship between emotional abuse and ADV perpetration for males, as well as the relationship between emotional abuse or physical abuse and ADV victimisation for females. ADV perpetration was predicted by child maltreatment histories, with adolescent anger functioning as a mediator. In this context, trauma-related anger is, in part, explanatory for aggression against a partner; it should be noted, though, that some of this behaviour may reflect a violent dynamic of back-and-forth, potentially escalating aggression, as the CADRI does not capture the interplay but only presence or frequency. Considering ADV perpetration, exposure to caregiver IPV was a predictor, indirectly, *via* trauma-related anxiety and anger (75).

Intellectual disabilities and adolescent attachment styles also played a role in ADV. Attachment style is conceptualised as a generally consistent variable over time and was measured by Attachment Security Ratings (86). Avoidant attachment denotes discomfort towards long-term relationships (87), and an avoidant style reflects a tendency to avoid emotional closeness. Thus, both intellectual disability and attachment style would be historical, although the attachment style in adolescence is not crystallised and would be expected to change, for example, with intervention (86). Adolescents with borderline to mild intellectual disabilities reported significantly more experiences of ADV victimisation and perpetration, compared to adolescents with average IQ. Particularly among adolescents with lower IQ, having an avoidant attachment style significantly predicted both ADV victimisation and perpetration, indicating the risk in avoidance as a style for interacting with others (78). The secure attachment style reflects flexible responding, and confidence in the other interactant to provide a secure base for development, support, and exploration. An avoidant interpersonal style may be protective in terms of exposure to dangerous persons and situations; however, it represents a limitation in experiencing emotional connectedness.

Substance Use

Histories of child maltreatment, trauma symptoms (i.e., anger, anxiety, dissociation, and sexual concerns), and Indigenous identity had significant impacts on substance use among MAP study participants. Greater alcohol problems were predicted by child maltreatment histories and were mediated by adolescent anger (75). In the same sample, 38.6% of youth reported at least one incident of binge drinking in the last 30 days, 40.3% of youth reported alcohol use in the past year, and 10.1% of youth met the cut-off for the development of a drinking problem. Given that the legal drinking age in Ontario (age 19) was outside of all MAP study participants, these numbers are concerning. In Hudson et al. (81), CSA history was associated with problem drinking and was partially mediated by anger for male adolescents, and fully mediated by anxiety and anger for female adolescents.

A significant number of MAP youth reported drug use. Faulkner et al. (75) found that 45.6% of the sample reported cannabis use in the last 30 days. In addition, 19.3% of youth in child welfare reported using illicit drugs. Similarly, child maltreatment history was significantly and positively associated with alcohol problems (79). In Goldstein et al. (80), anger and dissociation symptoms were positively associated with alcohol and drug problems (80). Child maltreatment histories and dissociation were both positively associated with using a greater number of illicit drugs in the past year (80). With regards to cannabis usage specifically, associations were found with exposure to caregiver IPV, mediated by anxiety, dissociation, and anger (75).

Cannabis usage was not dependent on Indigenous identity, as Indigenous and non-Indigenous youth reported no differences in Waechter et al. (84). However, Indigenous youth who reported low positive identity with their caseworker were 5.47 times more likely to have used cannabis in the past 12 months, compared to non-Indigenous youth with a low caseworker identification (84).

Sexual Risk-Taking

Sexual health and sexual risk-taking are interconnected. Early entry into sexual relationships was notable for MAP males (25% before age 13), and sexual health practises varied (e.g., 12% of MAP females reported that they never used protection in intercourse) (85). Sexual risk-taking indicators formed a risk-taking score and were assessed as age at first sexual intercourse, alcohol/drug use before engaging in sex, condom use, positive testing for sexually transmitted infections, and a total number of sexual partners. Wekerle et al. (76) reported a significant overall relationship between CSA history and adolescent sexual risk-taking; for instance, adolescents with a history of CSA were more likely than those without a history to have had sex with multiple partners.

Body Function Impairments

Coping Motives

Coping motives predominantly acted as mediators for the relationships across child maltreatment histories, trauma symptoms, and alcohol misuse or sexual risk-taking. For example, coping motives (e.g., to become more sociable, comply with peer pressure, forget about worries, feel good) for drinking were significantly and positively associated with child maltreatment and anxiety (79). For those with high maladaptive coping motives, greater anxiety was associated with more alcohol problems, whereas lower depression was associated with fewer alcohol problems. In Park et al. (82), both trauma symptoms and coping motives mediated the positive relationship between child maltreatment and alcohol misuse *via* serial mediation (82), showing that both are important for understanding adolescent substance use.

Child sexual abuse (CSA) was also associated with a greater number of maladaptive coping motives, as compared to non-CSA MAP study youth. CSA youth were significantly higher on having sex to cope with negative emotions, in both males and females than non-CSA youth. CSA history was also associated with greater maladaptive coping motives for sex (i.e., having sex in order to gain peer and/or partner approval), which in turn were associated with increased sexual risk-taking (i.e., a higher number of partners, not using protection during sex, using alcohol or drugs before sex) (76).

Sleep Disturbances

Sleep disturbances in the past 30 days were analysed by one study and measured with 11 self-report questions adapted from standardised questionnaires. McPhie et al. (51) found that the most reported sleep problems include taking longer than 30 min to fall asleep (61.6%), waking up before intended (46.4%), and having non-restorative sleep (38.4%). It was also reported that the severity of childhood maltreatment was predictive of sleep disturbances, where psychological distress functioned as a mediator. As the distress measure tapped proximal distress, it suggests that such issues as trauma symptoms, anxiety, and anger, as previously noted, are current candidates in determining body or physical regulation.

DISCUSSION

Multiple outcome research from a large child welfare system dataset with multiple types of mental health and functioning measures support the utility of an ICF approach to understanding the holistic health of maltreated youth who come to the attention of formal child welfare systems. An ICF approach provides added value in directing assessment attention (e.g., adolescent dating violence, sexual risk-taking), in an ongoing fashion, and incorporates domains of clinical relevance (e.g., coping motives), not typically included in assessment models. As indicated in this study, unique and significant challenges are faced by youth in child welfare settings. A distal vs. proximal model seems important to understand the interplay of factors and the changing landscapes of maltreated adolescents receiving child welfare services. Histories of child maltreatment were consistently and positively correlated with mental and physical health conditions (i.e., trauma symptoms, intellectual disabilities) and functioning problems (i.e., substance use, ADV, sleep problems, sexual risk-taking). For example, all maltreatment types and trauma symptoms presented positive relationships, showing the important linkages and mediator role of trauma symptomatology and, hence, the need to assess adolescent trauma symptomatology. In terms of specific types, exposure, and certain maltreatment forms (IPV, CSA, and emotional abuse) emerged as impacting functioning more significantly. Histories of emotional abuse were predictive of both PTSS and ADV in the past year for male and female youth (85), yet, relatively little attention is given to it clinically (88). The practitioner under-attention to emotional or psychological maltreatment prompted a statement from the American Paediatrics Association to review findings and identify that it needs to be considered alongside and independently from the more often considered physical and sexual abuse, in terms of assessment, prevention, and the potential utility of trauma-focused treatments (89).

While the popular CTQ measure was primarily used for investigating historical child maltreatment in the MAP sub-studies, the CEVQ filled the gap of providing information on exposure to IPV. The historical childhood exposure to IPV as a distal factor was significantly linked to the proximal experience of ADV, in which trauma symptomatology emerges as a bridging factor. ADV emerged differently over time, such that at a mid-adolescent timepoint (i.e., the MAP initial age described youth, on average, ranging from 15 to 16 years), the minority of youth reported ADV. However, with a view from the initial assessment to the 2-year follow-up, where youth are in late adolescence (ages 17–18 years), most youth had experienced some form of ADV. High rates of ADV perpetration and victimisation indicate the need for interventions on relationship skill building and safety skills. Foster care and non-foster care youth vary in their access to resources, so making sure these interventions are equitably distributed is essential. This is especially important when considering the increased prevalence of ADV victimisation among non-foster care male youth, given that greater service provision typically is directed to foster care youth (83). Thus, when it comes to relationship violence, a distal by proximal view seems important and needs to be part of the ongoing inquiry during caseworker visits and in adolescent medicine.

The ICF framework accommodates well the interplay between environmental factors with health conditions. Given the placement disruptions of youth in the MAP study, the risk for disrupted attachments, trauma, and instability in social networks seem more likely. Analyses of child welfare statistics in Canada show that youth in care face increased environmental turbulence, such as multiple placement moves (90). A greater number of moves has been associated with difficulties in building consistent relationships in new schools and community neighbourhood settings, as well as the development of more severe trauma symptoms (91, 92). Practitioners are encouraged to continue checking such quality-of-life factors, particularly for those involved in the child welfare system. Given the consistent mediator role of trauma-related emotionality (distress, anxiety, anger) for both males and females, supporting youth to be more fully engaged in the decision-making is not only a rights-based expectation (i.e., sustainable development goal 16, right to be free from all forms of violence) but also a principle of trauma-informed care (93). During adolescence, it is critical for youth to grow and develop in supportive environments, with ample opportunities to connect safely, access resources, and practise adaptive emotion regulation skills. Youth with histories of greater poly-victimisation are more likely to experience maltreatment while in foster care, compared to those with fewer maltreatment experiences (94), and care needs to be prioritised to prevent such re-victimisation and traumatization. The medical home may be an important point of continuity of care.

It was found that child welfare-involved youth with borderline-to-mild intellectual disabilities reported significantly greater dating violence victimisation and perpetration, compared to youth with average IQs (78). Children with disabilities are at greater risk of experiencing child maltreatment, with three times the higher prevalence of maltreatment than the typically developing population (95). When considering the whole child welfare system, ∼50% of children investigated by CPS had a developmental disability(ies) (95). More concerning is the understanding that maltreatment rates are likely higher than reported, as those with disabilities may experience communication difficulties that hinder disclosure or self-report to caseworkers or practitioners.

Weiss et al. (78) further detected the role of attachment style, where avoidant attachment styles correlated with both ADV victimisation and perpetration, particularly among adolescents with low IQ. Children with maltreatment histories are less likely to seek abusive attachment figures (e.g., parents, caregivers) for support or comfort, which can, in turn, lead to increased fears related to attachment figures and, hence, the development of avoidant attachment styles (96). Insecure attachment models, which begin at an early age, likely carry out into adolescence in terms of developing a stylistic way of relating which, in turn, can impact the ability to engage in healthy relationships (97). It may, therefore, be useful to consider the maltreated, child welfare-involved youth's experiences with close relationships, and how that influences their approach/avoidance styles. Youth with an avoidant style may not make the connexion to how this style of interaction expresses in a romantic relationship, for example, and understanding this dynamic may help to direct the clinician's anticipatory guidance approach. Research has shown the potential of modifying attachment styles *via* treatment and can therefore be a goal of intervention for child welfare-involved youth, who are at greater risk for ADV involvement (98).

Child maltreatment experiences, trauma symptoms, and coping motives were frequently linked together in the MAP sub-studies. Emotion regulation emerges as a core issue in understanding both the tendency towards emotion avoidance (e.g., avoidant attachment style) and the mediation by trauma-related symptomatology. The MAP sub-study findings suggest a coherence in the core issue of emotion regulation and coping across diverse outcomes (i.e., ADV, substance use, or sexual risk-taking). Substance use across child welfare settings stood out as a particular concern. Similar to childhood IPV exposure and later dating violence, the social learning theory argues how youth can learn positive or negative behaviours from observing others that inform the experiences of victimisation and later coping strategies (99). If substance use or IPV is prevalent in initial households, as these tend to overlap [e.g., (100)], this could be a contributing factor to later substance use and ADV problems. Indeed, MAP youth with CSA histories identified motivations to have sex as focused on a way to cope with negative affect. Alternatives to coping with trauma-related negative affect and addressing emotion regulation challenges (e.g., emotional reactivity to threat/perceived threat, delayed emotional responding with dissociation) may be an important clinical goal. These higher rates of substance use among MAP youths are consistent with previous studies. For example, youth with four or more adverse childhood experiences are approximately two, seven, five, and 11 times more likely to engage in smoking, alcoholism, illicit drug use, and injected drug use, respectively, than those with fewer ACEs (101). Substance use was also often associated with coping motives, which we considered an impaired body function. Drinking, for example, may serve as a tension-reduction mechanism to cope with feelings of anxiety and, in turn, better allow youth to participate in social events in dampening social anxiety. Such coping may work in tandem with the impact of child maltreatment histories, which are also associated with the inability to develop healthy coping motives. These coping motives were also present in reasons for having sex, particularly among those with CSA histories. Adaptive coping and social skill development are important targets to consider for child welfare system-involved youths.

Clinical Implications

For the practitioner, the ICF model is valuable in guiding service provision to foster care and/or maltreated youth. Currently, the HEADSS (Home; Education and employment, eating and exercise; Activities and peer relationships, social media; Drug use, including prescribed medications, cigarettes, vaping, alcohol and other drugs; Sexuality and gender; Suicide, self-harm, safety, and spirituality) assessment is used as a psychosocial assessment for adolescents by health practitioners (102). In reference to child welfare-involved youth, however, it is a missed opportunity to understand the why behind health-risk behaviours, such as maladaptive coping. For example, coping motives were significant in correlating with alcohol problems and sexual risk-taking behaviour (79, 82). Both behaviours are being engaged in for managing negative emotions, which highlights the need for services that directly target the management of emotions (i.e., emotion literacy, adaptive emotion expression, managing distress, alternatives to acting-out behaviours, and resilience strategies). Interventions that support new routines in emotion regulation would seem important, such as Trauma-focused Cognitive Behaviour Therapy, which addresses PTSS with cognitive-behavioural principles (103); and interventions based on behavioural perspectives, which focuses on understanding antecedents and consequences of actions (104). More research is needed to determine which interventions are most effective for child welfare-involved youth particularly. The interrelated nature of the ICF model further reinforces the ways in which contextual factors and health conditions interact to influence functioning, appropriate to the complex nature of youth experiences within child welfare.

Additionally, it may be practical to consider the "medical home" when providing care for child welfare-involved youth (105). A medical home is an approach that integrates patients, families, clinicians, and medical staff, to provide comprehensive primary care. They are well-known in their ability to conduct ongoing screening and facilitate longitudinal relationships, which are critical for the turbulent environment of child welfare-involved youth. The medical home can stand as a source of stability. Research has shown that youth with special healthcare needs (i.e., requiring additional health-related services due to increased risk of developmental, physical, emotional, and/or behavioural conditions) benefit from the medical home, as seen by improvements in health, school, and work attendance, as well as access to care (106). The medical home, as an environmental factor may, therefore, facilitate functioning in this population. In a meta-analysis, it has also been found that communication with parents about safe sex can act as a protective role in adolescent safer sex behaviour (107). Professionals in the medical home, or caregivers in the child welfare system, can have these conversations or support guardians in this regard, as prevention against sexual risk-taking and compromised sexual health.

It is a given that clinicians interacting with youth from the child welfare system be well-versed in current literature in the child maltreatment area. For developing clinicians and trainees, various resources and guidelines currently exist for clinicians to use in the care for child welfare-involved youth, including The Encyclopaedia of Early Childhood Development (108) and the VEGA (Violence, Evidence, Guidance, Action) Project (109), as open access options for information and training about detecting and responding appropriately to child maltreatment and family violence. Similarly, there are several guidelines and tools for mental health treatment, such as the WHO Mental Health Gap Action Program (110) and the American Academy of Paediatrics primary care tools (111). Using such tools can support better care for vulnerable youth.

Strengths and Limitations

Strengths of this review include the use of the ICF to analyze the health and functioning of child welfare-involved youth. As seen in the MAP sub-studies, multiple factors were connected and significantly related to one another. Trauma symptoms often functioned as mediators (e.g., relationships between sleep problems and child maltreatment histories; exposure to IPV and substance use, etc.), which aligns with the multi-faceted nature of the ICF model.

Limitations include the incompatibility between the measure timelines and the ICF model. The ICF is a snapshot, meaning it represents the functioning of an individual at a specific time; comparatively, the MAP study measures included a wide range of timeframes. For example, child maltreatment was measured by the CTQ-SF, which reports maltreatment history "while growing up" without a specific time frame; in contrast, the CEVQ reports the frequency of IPV and maltreatment experiences more specifically (e.g., between grades 1–5, grades 6–8, happening now, etc.) providing better quantification of distal vs. proximal influences. Moving forward, questionnaires should be standardised in a way that can disentangle proximal vs. distal time points, and graphical databases may be implemented to visualise the various timelines.

The MAP sub-studies were also ambiguous in using the terms, "gender" and "sex," interchangeably. This may be attributed to the publication year of some studies, during a time in which definitions of gender and sex definitions were not properly delineated. While sex was analysed in the MAP sub-studies, it can be insightful to expand research to include a range of genders, as gender and sexual minorities may be at greater risk of adverse outcomes in child welfare settings (49). Similarly, future steps should involve further analysis of the disproportionate representation of Indigenous youth in child welfare (112). Family and community may both be significant factors of resilience in Indigenous youth, who strongly value and uphold relationships, which can better contribute to appropriate activities and participation in daily life. The MAP study was limited in its analysis of this population, but even then, was able to detect the importance of relationships, as Indigenous youth who experienced good caseworker relationships were significantly less likely to engage in cannabis use. In Ontario, if a youth self-identifies as Indigenous, they are directed to the Indigenous child welfare agency, if available in that locale. The over-representation of diversity sub-groups, and its potential overlap with socioeconomic disadvantage, is an important ongoing area that child welfare continues to contend with, and is addressed with different service models.

CONCLUSIONS

The environment in which a youth is embedded is critical to their ability to function and navigate life in a healthy manner. This is even more true for youth in child welfare settings, experiencing a high rate of adversity and ongoing risk. Any interventions, or solutions, to promote the well-being of child welfare-involved youth ought to address the various interrelated factors involved, which the ICF framework is well-suited to achieve.

AUTHOR CONTRIBUTIONS

KK, CM, JP, and CW contributed to conceptualising the review topic and aim. KK, CM, and JP extracted and analysed data. KK wrote the majority of the first manuscript draft. All authors contributed to writing parts of the manuscript, provided feedback on an ongoing basis, revised the manuscript, and approved the submitted version.

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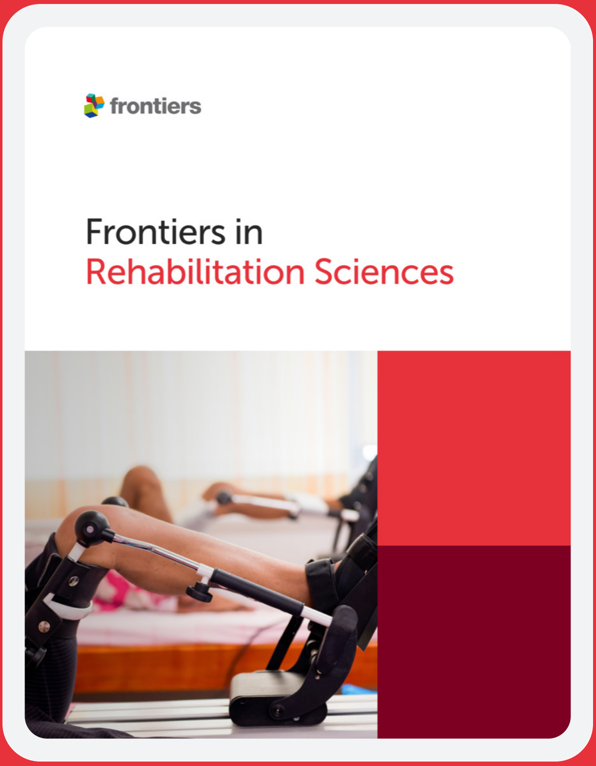

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