Parental and Medical Classification of Neurodevelopment in Children Born Preterm

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abstract

BACKGROUND AND OBJECTIVES: The likelihood and severity of neurodevelopmental impairment (NDI) affects critical health care decisions. NDI definitions were developed without parental perspectives. We investigated the agreement between parental vs medical classification of NDI among children born preterm.

METHODS: In this multicenter study, parents of children born preterm (<29 weeks) evaluated at 18 to 21 months corrected age (CA) were asked whether they considered their child as developing normally, having mild/moderate impairment, or having severe impairment. Medical categorization was based on hearing, vision, cerebral palsy status, and Bayley Scales of Infant and Toddler Development Third Edition (Bayley-III) scores. Agreement was analyzed using Cohen's weighted κ. Discrepancies in categorization by NDI components and parental demographics were examined using the Pearson χ^2 test, Fisher exact test, or Wilcoxon signed-rank test.

RESULTS: Children (n = 1098, gestational age 26.1 ± 1.5 weeks, birthweight 919 ± 247 g) were evaluated at 19.6 ± 2.6 months CA at 13 clinics. Agreement between parental and medical NDI classification was poor (κ = 0.30; 95% CI: 0.26–0.35). Parents described their child's development as normal or less impaired. Only 12% of parents of children classified as having a severe NDI according to the medical definition agreed. There were significant disagreements between classification for children based on Bayley-III cognitive, language, and motor scores but not for cerebral palsy. Discrepancies varied by parental education and ethnicity but not by single caregiver status.

CONCLUSIONS: Parent perception of NDI differs from medical categorization, creating a risk of miscommunication. This indicates an overestimation of the impact of disability by clinicians, which may affect life-and-death decisions. Parental perspectives should be considered when reporting and discussing neurodevelopmental outcomes.

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WHAT'S KNOWN ON THIS SUBJECT: In cases of extreme prematurity, the likelihood and severity of neurodevelopmental impairment (NDI) affects critical health care decisions. Medical definitions of NDI were developed without parental perspectives. It is unknown whether parents of children born preterm and clinicians/researchers agree on classifications of NDI.

WHAT THIS STUDY ADDS: In preterm outcome studies, reported levels of impairment have been chosen by clinicians/researchers. Parent perception of severity of NDI differs from published data, creating a risk of miscommunication. Future studies should revise outcome definitions to incorporate parental values and priorities.

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INTRODUCTION

Children born preterm (<37 weeks' gestational age [GA]) are more likely to have health and neurodevelopmental challenges.[1,2] Their parents often have concerns about the future, and clinicians answer their questions about the outcomes of prematurity using findings from clinical follow-up audits and population-based studies.[3] Outcomes studies are used to counsel women experiencing preterm labor, provide prognostic information, and make decisions about life-sustaining interventions.[4–8] Preterm birth outcome studies and clinical follow-up have traditionally focused on neurodevelopmental impairment (NDI) and use outcome classification and definitions that were developed by researchers and clinicians—without input from parents.[9–11]

Population-based perinatal networks with ongoing neurodevelopmental surveillance programs of high-risk infants, such as those born extremely preterm, have standardized assessments between 18 and 36 months corrected age (CA). This is often composed of a neurological examination to identify signs of cerebral palsy (CP), identifying the degree of functional impact using the Gross Motor Function Classification System (GMFCS), a standardized developmental assessment using the Bayley Scales of Infant and Toddler Development Third Edition (Bayley-III), and determination of hearing and visual function. Similar to many neonatal follow-up programs around the world, infants born preterm before 29 weeks' GA in Canada are invited to have a standardized health and neurodevelopmental assessment at 18 to 21 months CA.[12] The Canadian Neonatal Follow-Up Network (CNFUN) has published studies and reports using the composite outcomes based on the components listed above, and outcomes are often classified according to levels of NDI: none, mild/moderate, or severe NDI.[13] Internationally, definitions of NDI and severity level may vary from one setting to the other with no clear consensus on measuring and reporting.[14] Moreover, parents have never been asked whether this classification was accurate or meaningful to them.

To address this knowledge gap, we conducted a national multicenter cross-sectional study investigating parental perception of their own child's level of neurodevelopment at 18 to 21 months CA in comparison to the CNFUN medical classification of NDI.

METHODS

Study Population

This is a cross-sectional observational study in which all parents/caregivers of children born before 29 weeks GA and seen at the 18- to 21-month CA follow-up visit at a participating CNFUN site from May 2017 until September 2021 were eligible to participate. This study was approved by the University of British Columbia Children's & Women's Research Ethics Board (H17–03490). Participating sites obtained approval at their individual hospital research ethics board. Consent was obtained when required by institutional research ethics boards. Sites were excluded if they submitted parent classification data for less than 50% of patients seen starting from when they obtained ethics approval at their individual site.

Procedure

At the CNFUN visit, a half-page information document (cocreated with parent-partners) described the purpose of the neurodevelopmental assessment, which is to provide up to date information on outcomes of children born extremely preterm that can be used for clinical care, benchmarking, and research (Supplement 1). Prior to the standardized evaluation, parents were asked the following question: "Please tell us how you would rate your child's development: (1) my child is developing normally, (2) my child has a mild developmental impairment, (3) my child has a moderate developmental impairment, or (4) my child has a severe developmental impairment." Responses were recorded and linked to the child's health information from the CNFUN database using their unique neonatal follow-up identification number. The CNFUN visit assessment proceeded as per the standard protocol.[15] We obtained general demographic information, GA at birth, birth weight, age, date when visit occurred, parental demographic information, and results from the medical assessment from the CNFUN database.[12]

Outcome Measures

The primary outcome was the agreement between parents' classification of their child's overall NDI status and the CNFUN classification. As per CNFUN classification, a child is considered to have a mild-moderate NDI if they had any 1 or more of the following: CP with GMFCS 1 or 2; Bayley-III motor, cognitive, or language composite scores 70 to 84; hearing loss without requirement for hearing devices or unilateral visual impairment. A child is considered to have a severe NDI if they had any 1 or more of the following: CP with GMFCS 3, 4, or 5; Bayley-III motor, cognitive or language composite scores <70; hearing aid or cochlear implant; or bilateral visual impairment. Exploratory analyses were conducted to identify factors associated with discrepancies between parental and CNFUN classifications.

Statistical Analysis

The proportion of children in each of the categories by parental classification and CNFUN NDI classification were analyzed descriptively and cross-tabulated. Parental classification of mild and moderate developmental impairment was combined into 1 category to align with the CNFUN NDI category of mild to moderate impairment. Cohen's kappa coefficient (κ) was chosen as the optimal quantitative

measure of reliability for 2 participants who were rating the same concept. First, Cohen's κ was analyzed to determine chance independent agreement between parental and CNFUN classification. Second, Cohen's weighted κ was analyzed to determine agreement between parental and CNFUN categorizations, considering the closeness of agreement between categories (ie, no NDI, mild/moderate NDI, severe NDI). We determined the acceptability of κ where <0.40 = poor, 0.40 to 0.59 = fair, 0.60 to 0.74 = good, and 0.75 to 1.00 = excellent.[16] κ less than 0.60 (poor or fair) was judged as being unacceptable when comparing NDI in the context of neonatal follow-up. We examined demographic and clinical factors associated with discrepancies between parental and CNFUN categorizations using the Pearson χ^2 test or Fisher exact test for categorical variables and the Wilcoxon signed-rank test for continuous variables. To assess factors associated with disagreements, we used multivariate regression modeling adjusted for potential confounders to obtain adjusted odds ratios and 95% CIs. The generalized estimated equation was used to adjust for clustering within multiples and by site. If cell size for a characteristic was less than 5, these small values were not reported to maintain patient confidentiality. All analyses were conducted using SAS 9.4 (SAS Institute Inc, Cary, NC).

RESULTS

Among 13 participating CNFUN sites in this study, 1386 patients were seen at the 18- to 21-month CA visit. Children seen at follow-up were born at earlier GA, had lower birth weight, were more likely to have received antenatal corticosteroids, and more likely to have bronchopulmonary dysplasia (BPD) than children who did not attend the visit. Among these, 1098 parents (79%) of children born extremely preterm participated.

Demographic characteristics were similar between participating and nonparticipating families (Table 1). Children of participants were born at an average of 26.1 weeks, with a birth weight of 919 g, and were assessed at 19.6 months CA. Following neurodevelopmental assessment, 495 (45%) were identified as having at least 1 impairment. Median Bayley-III cognitive, language, and motor composite scores were 95, 89, and 94, respectively. Parent participants included 55 (5%) single caregivers and 921 (92%) with completed postsecondary education.

Agreement of Parental Classification vs CNFUN NDI Classification

When comparing the chance independent agreement between parental vs CNFUN classification of NDI using both Cohen's simple κ and weighted κ , there was poor agreement (κ = 0.24; 95% CI: 0.20–0.29 and κ = 0.30; 95% CI: 0.26–0.35, respectively). Parents were more likely to describe their child's development as normal or less impaired than the CNFUN classifications (Table 2). According to the CNFUN classification, 54%, 29%, and 17% of children were classified as having no NDI, mild-moderate NDI, or severe NDI, respectively (vs 67%, 31%, and 2% by parental classification). For 59% of children, parents and CNFUN agreed on NDI classification, and for 31%, parents rated their child's neurodevelopment more positively than the CNFUN classification (Table 2). Among the 596 children who were considered to have no NDI, 491 (82%) were also perceived as such by parents. Among those with mild-moderate NDI as per CNFUN, agreement with parents was 42%. Among the 185 children with severe NDI according to CNFUN, only 23 (12%) received a similar rating by their parents.

Disagreement Between Parental and CNFUN Classification for Severe NDI

Children who were classified as having a severe NDI by CNFUN (n = 185) were most often considered to have a mild-moderate NDI by parents (n = 103). In addition, almost a third (n = 59) were rated as having no NDI by their parents. When there was disagreement, these children typically displayed higher Bayley-III Cognitive and Motor Composite scores (Table 3). Parents were more likely to agree with the CNFUN classification of severe NDI when the child had a physician's rating of severe global developmental delay.

Disagreement Between Parental and CNFUN Classification for Mild-Moderate NDI

Children who were classified as having a mild-moderate NDI by CNFUN but were classified as having no NDI by parents had higher Bayley-III language and motor composite scores compared to those in whom there was agreement (Table 4). There was agreement in classification for children with CP or hearing or visual impairments. Parents of children who required health care resources since neonatal intensive care unit (NICU) discharge including rehospitalization, technology used in the home, or referral to services were more likely to agree with the CNFUN classification for mild-moderate NDI.

Disagreement Between Parental and CNFUN Classification for No NDI

Children who were classified as having no NDI by CNFUN but were classified as having a mild-moderate or severe NDI by parents had lower Bayley-III language and motor composite scores (Table 5). Parents were more likely to disagree with the CNFUN classification of no NDI when the child required use of any technology in the home such as supplemental oxygen, feeding tubes, or mobility aids or received any referral since NICU discharge to an occupational therapist, physical therapist, psychologist, rehabilitation program, or speech/language therapist.

TABLE 1. Characteristics of Participating and Nonparticipating Families at Canadian Neonatal Follow-Up Network Participating Sites

Characteristics	Study Participants, n = 1098	Nonparticipants, n = 288	P Value
Birth and child information			
Gestational age at birth, mean (SD), wk	26.1 (1.5)	26.1 (1.6)	.99
Birth weight, mean (SD), g	919 (247)	895 (258)	.14
Corrected age when visit occurred, mean (SD), mo	19.6 (2.6)	19.7 (2.7)	.48
Neurodevelopmental assessment			
Bayley-III cognitive composite score, median (IQR)	95 (90–105)	95 (85–100)	**.01**
Bayley-III language composite score, median (IQR)	89 (77–100)	89 (77–100)	.46
Bayley-III motor composite score, median (IQR)	94 (85–100)	94 (85–100)	.44
Definitive CP, n (%)	65 (6.0)	18 (6.5)	.75
CP with GMFCS I or II, n (%)	45 (71.4)	8 (53.3)	.18
CP with GMFCS III, IV, or V, n (%)	18 (28.6)	7 (46.7)	
Hearing impairment: sensorineural/mixed hearing loss not requiring aid, n (%)	64 (6.0)	14 (5.1)	.58
Hearing impairment: hearing aid or cochlear implant, n (%)	13 (1.2)	<5	.99
Visual impairment: unilateral, n (%)	<5	<5	.99
Visual impairment: bilateral, n (%)	9 (0.9)	<5	.72
No impairment, n (%)	603 (54.9)	151 (52.4)	.71
Any 1 impairment, n (%)	280 (25.5)	81 (28.1)	
Multiple impairments: any 2 impairments, n (%)	118 (10.8)	28 (9.7)	
Multiple impairments: any 3 impairments, n (%)	68 (6.2)	22 (7.6)	
Multiple impairments: any 4 impairments, n (%)	25 (2.3)	6 (2.1)	
If no Bayley-III scores, clinician's rating as severe global developmental delay, n (%)	23 (21.7)	16 (53.3)	**<.01**
Parent demographics			
Single caregivers, n (%)	55 (5.1)	21 (7.5)	.11
2 caregivers, n (%)	1033 (94.9)	260 (92.5)	
Education: completed postsecondary or higher, n (%)	921 (91.6)	231 (87.8)	.06
Education: incomplete postsecondary or lower, n (%)	84 (8.4)	32 (12.2)	
1 employed caregiver (of children with 1 caregiver), n (%)	28 (50.9)	11 (52.4)	.91
2 employed caregivers (of children with 2 caregivers), n (%)	585 (56.6)	144 (55.4)	.72
Self-reported ethnic group: white, n (%)	507 (49.1)	106 (43.4)	.07
Self-reported ethnic group: Black, n (%)	114 (11.1)	41 (16.8)	
Self-reported ethnic group: Asian, n (%)	232 (22.5)	55 (22.5)	
Self-reported ethnic group: Hispanic/Latino, n (%)	33 (3.2)	6 (2.5)	
Self-reported ethnic group: Indigenous, n (%)	37 (3.6)	12 (4.9)	
Self-reported ethnic group: unspecified, n (%)	40 (3.9)	<5	
Self-reported ethnic group: unreported, n (%)	69 (6.7)	20 (8.2)	
Resource use			
Any rehospitalization, n (%)	404 (36.9)	92 (32.9)	.21
Use of any technology in the home, n (%)	96 (8.7)	29 (10.2)	.44
Any referral to occupational therapist, physical therapist, psychologist, rehabilitation program, or speech/language therapy	777 (84.8)	172 (80.0)	.08
Any referral to occupational therapist	465 (52.7)	101 (51.8)	.81
Any referral to physical therapist	572 (65.2)	105 (53.0)	**<.01**
Any referral to psychologist	63 (7.6)	11 (5.8)	.39
Any referral to rehabilitation program	122 (14.5)	38 (19.8)	.07
Any referral to speech/language therapy	414 (47.2)	111 (54.2)	.07

Abbreviations: IQR, interquartile range; CP, cerebral palsy; Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition; CNFUN, Canadian Neonatal Follow-Up Network; GMFCS, Gross Motor Function Classification System.
Participating site is defined as a CNFUN site which submitted parent classification data for at least 50% of patients seen. The timeframe of patients seen started from the first entry upon obtaining ethics approval until the last entry of parent classification data (active contribution) at that individual site. Bold indicates significance.

Discrepancies in Classification by Parent Demographics

Discrepancies varied by parental education and ethnicity but not by single caregiver status. Parents who completed postsecondary education and who reported being of white ethnicity were more likely to agree with CNFUN categorization (58% vs 46% and 63% vs 55%, respectively; Supplemental Table 1).

Factors Associated With Disagreement

After adjustment for confounders, the factors found to be associated with disagreement were Bayley-III cognitive and language composite scores as well as self-reported ethnicity (Supplemental Table 2).

TABLE 2. Agreement of Parental Classification and CNFUN NDI Classification

		CNFUN Classification			
		No NDI	Mild-Moderate NDI	Severe NDI	Total, n (%)
Parental Classification	No NDI	491	182	59	732 (67)
	Mild-Moderate NDI	104	131	103	338 (31)
	Severe NDI	11	2	23	26 (2)
Total		596 (54)	315 (29)	185 (17)	1096

Abbreviations: CNFUN, Canadian Neonatal Follow-Up Network; NDI, neurodevelopmental impairment.

TABLE 3. Understanding Differences for CNFUN Severe NDI Classification

NDI Domain	CNFUN Severe NDI Agreement, n = 23	CNFUN Severe NDI vs Mild-Moderate NDI Classification by Parents, n = 103	*P* Value	CNFUN Severe NDI vs No NDI Classification by Parents, n = 59	*P* Value
Bayley-III cognitive, median (IQR)	70 (60–85)	80 (65–85)	0.23	80 (75–95)	**0.04**
Bayley-III language, median (IQR)	59 (52–67)	64 (56–68)	0.28	62 (56–68)	0.36
Bayley-III motor, median (IQR)	55 (49–70)	73 (61–88)	**0.01**	82 (67–94)	<**0.01**
CP: GMFCS I–II, n (%)	<5	8 (34.8)	**0.04**	<5	0.27
CP: GMFCS III, n (%)	<5	8 (34.8)		<5	
CP: GMFCS IV–V, n (%)	11 (73.3)	7 (30.4)		<5	
Hearing impairment: hearing aid or cochlear implant, n (%)	<5	9 (8.7)	0.36	<5	0.57
Visual impairment: bilateral, n (%)	<5	6 (6.0)	0.17	<5	**0.02**
Clinician's rating as severe global developmental delay, n (%)	12 (52.2)	9 (8.7)	<**0.01**	<5	<**0.01**
Any rehospitalization, n (%)	15 (65.2)	53 (51.5)	0.23	26 (44.1)	0.09
Use of any technology in the home, n (%)	9 (39.1)	28 (27.2)	0.26	8 (13.6)	**0.02**
Any referral to occupational therapist, physical therapist, psychologist, rehabilitation program, or speech/language therapy, n (%)	23 (100)	93 (93.9)	0.59	51 (96.2)	1.00

Abbreviations: IQR, interquartile range; CP, cerebral palsy; Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition; CNFUN, Canadian Neonatal Follow-Up Network; GMFCS, Gross Motor Function Classification System; NDI, neurodevelopmental impairment.
P value compares group with agreement (column 2) vs disagreement (column 3 or 5). Bold indicates significance.

DISCUSSION

In this national multicenter study investigating the agreement between more than 1000 parents' perception of their extremely preterm child's level of NDI and the CNFUN classification, and clinicians' and researchers' definition of severe NDI differed from families' perceptions. This has significant implications, as perinatal studies often report rates of severe NDI as a primary outcome. This can then influence practices and, most importantly, decision-making for clinicians and parents, such as whether to withhold or withdraw life-sustaining interventions. Our study highlights the disagreement between families and research-based definitions of NDI and lends support to justify reframing the current classification system.

The lack of consensus on measuring and reporting NDI has led to controversy and challenges when interpreting outcomes in the literature. Severe NDI is typically reported as a composite outcome, but this definition has been shown to vary significantly across different perinatal networks. For example, Haslam et al identified 7 distinct definitions for severe NDI among 8 neonatal networks and reported a clinically important 4-fold difference in the incidence rates depending on the definition of severe NDI applied to the same population.[14] A similar definition used by CNFUN and the Vermont Oxford Network was the least stringent (resulting in highest incidence rate) compared to the other 6 networks.[14] The most stringent definition used by the Australia New Zealand Neonatal Network included at least 1 of the following: CP with GMFCS level 4 to 5, Bayley-III language, cognitive or motor composite score <3 SDs below the mean, or bilateral visual impairment.[17] There is no clear alignment on current definitions across networks, and our study provides new evidence that parents' perceptions are also not aligned with how outcomes are defined and reported.

Significant interest in measuring what is considered "normal" (defined by clinicians/researchers as a child without neurodevelopmental sequalae) has existed since the earliest publications of neonatal follow-up studies, dating back to the 1940s.[18] The largest and most rigorous study of infant outcomes in the 1960s, the Collaborative Perinatal Study, was organized by the National Institute of Neurological Diseases and Blindness, which shaped the trajectory toward neurodevelopment as a primary outcome of interest.[3] Despite variations in definition, NDI among survivors

TABLE 4. Understanding Differences for CNFUN Mild-Moderate NDI Classification

NDI Domain	CNFUN Mild-Moderate NDI Agreement, n = 131	CNFUN Mild-Moderate NDI vs No NDI Classification by Parents, n = 182	*P* Value
Bayley-III cognitive, median (IQR)	90 (85–95)	95 (85–100)	.09
Bayley-III language, median (IQR)	79 (74–89)	79 (77–89)	**.02**
Bayley-III motor, median (IQR)	85 (79–91)	94 (85–100)	**<.01**
CP: GMFCS I, n (%)	11 (68.7)	5 (83.3)	.78
CP: GMFCS II, n (%)	5 (31.3)	<5	
Hearing impairment: sensorineural/mixed hearing loss not requiring aid, n (%)	15 (11.7)	29 (16.2)	.27
Visual impairment: Unilateral, n (%)	<5	<5	.51
Any rehospitalization, n (%)	58 (44.3)	60 (33.0)	**.04**
Use of any technology in the home, n (%)	23 (17.6)	7 (3.9)	**<.01**
Any referral to occupational therapist, physical therapist, psychologist, rehabilitation program, or speech/language therapy, n (%)	123 (96.1)	123 (82.6)	**<.01**

Abbreviations: IQR, interquartile range; CP, cerebral palsy; Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition; CNFUN, Canadian Neonatal Follow-Up Network; GMFCS, Gross Motor Function Classification System; NDI, neurodevelopmental impairment.
Bold indicates significance.

TABLE 5. Understanding Differences for CNFUN No NDI Classification

NDI Domain	CNFUN No NDI Agreement, n = 491	CNFUN No NDI vs Mild-Moderate NDI Classification by Parents, n = 104	*P* Value
Bayley-III cognitive, median (IQR)	105 (95–110)	100 (95–110)	.55
Bayley-III language, median (IQR)	100 (91–109)	96 (89–100)	**<.01**
Bayley-III motor, median (IQR)	100 (94–107)	97 (94–100)	**.04**
Any rehospitalization, n (%)	151 (30.8)	39 (37.9)	.16
Use of any technology in the home, n (%)	10 (2.0)	10 (9.6)	**<.01**
Any referral to occupational therapist, physical therapist, psychologist, rehabilitation program, or speech/language therapy	275 (74.3)	87 (94.6)	**<.01**

Abbreviations: IQR, interquartile range; Bayley-III, Bayley Scales of Infant and Toddler Development, Third Edition; CNFUN, Canadian Neonatal Follow-Up Network; NDI, neurodevelopmental impairment.
Bold indicates significance.

measured at approximately 2 years of age has largely been adopted as the gold standard outcome in neonatal studies. This serves as a research endpoint for benchmarking neonatal care.[5,19] The choice to widely report severe NDI has important clinical implications because this information is used in prognostication, counseling, and clinical decision-making. For example, after a severe brain hemorrhage, many clinicians use data from follow-up studies, including the risk of "severe NDI" to guide discussions with parents about withholding or withdrawing life-sustaining interventions.[20] We have shown in this study that parent perception of severity of NDI differs from published data which creates a risk of miscommunication. This risk is particularly concerning during periods of critical decision-making.

Furthermore, parents' concerns in other domains (eg, behavior and feeding) are not included in the components of NDI.[21] Research has shown that parents value a more nuanced and balanced view of their child's health, function, and development rather than simplistic dichotomous categories (presence or absence of NDI).[21–23] A paradigm shift from a traditionally deficit-based approach in reported outcomes toward a strength-based functional approach has been proposed before by advocates, including parent-partners in research.[19] When compared to the CNFUN classification, we found that parents were more likely to describe their child's development favorably. Parents have been shown to rate their child's overall health as high, regardless of impairment.[22] Our study showed that despite a CNFUN classification of severe NDI, 88% of parents classified their child as having no or mild/moderate NDI. In addition, there are limitations to the use of these composite measures which assume each of its components bear equal weights. It has been shown that parents view the severity of the individual components of NDI defined by CNFUN differently.[24] Furthermore, there are ethical challenges of using composite outcomes for decision-making in the NICU, including how the presentation of information influences these decisions.[25] The appropriate denominator should be chosen during counseling; functioning would be optimal, or at least descriptions of impairment, as opposed to rates of "severe NDI" among survivors.

Although health care professionals have the knowledge and expertise to perform the assessments (neurological examination, the Bayley-III scales), to review audiology and ophthalmology reports, and to input this information in the CNFUN database, the classification of impairment in neonatal follow-up was done based on CNFUN definitions. However, parents are the ones who live with their children and are the experts on their own child's function and quality of life. Based on their 24/7 lived experience, parents may or may not describe their child as having a developmental disability.

The reasons why parents do not agree with the current NDI classification and severity may be multifactorial: (1)

adaptation and resilience, (2) parents value function and quality of life over diagnoses, and (3) 18 to 24 months CA may be too young for prognostication. Parents of children born extremely preterm have been shown to demonstrate resilience with respect to having an improved outlook on life and stronger family relationships despite increased vulnerability, stress, and a loss of personal and family equilibrium.[26] Most disagreement in our study occurred with Bayley-III scores, which have been shown to poorly predict long-term functioning, except for scores at the lowest extremes.[27–30] Parents are not in denial about their child's health status as they still express concerns regarding development and specific health issues, but parents likely perceive things differently than researchers.[21–23] Parents value their child's personality, happiness, function, and quality of life, regardless of the presence or absence of NDI, which may be factors equally relevant in their decision-making.[19,23] Parents acknowledge that NDI has an impact on their child's life, but other factors can also affect their quality of life.[31–34] Information on how the child is functioning and participating in daily activities is also vital to parents. Interestingly, we found that 105 children who were classified as having no NDI by CNFUN were classified as having some level of impairment by parents, which suggests that other important factors, such as the use of medical technology (eg, feeding tube, oxygen), were considered by parents when rating their child that are not captured in current definitions. Lastly, we found that parental demographics (ie, higher parental education and reported White ethnicity) influenced agreement. These factors may be more congruent with the current medical classification system; however, this highlights the need for more inclusive and equitable definitions to serve the diverse patient population seen in neonatal follow-up.

Although many studies have investigated the short- and long-term outcomes of prematurity,[35–37] no standardized reporting guidelines for neonatal follow-up exists. Past investigations have asked parents about a set of outcomes judged to be important to researchers and clinicians (using a top-down approach).[38,39] For example, parents were asked if BPD, necrotizing enterocolitis and CP were important as outcomes. On the other hand, recent bottom-up approaches have demonstrated that when parents are asked in their own words what is important, they prioritize function over diagnosis.[22–24,26,40–42] Instead of BPD diagnosis, other functional outcomes, such as leaving home with oxygen, were identified as important.[41] Furthermore, studies such as these will help to empower parents and families so they can advocate for their own child's needs.[19,43] In other medical fields, outcomes are reported without value, such as in oncology. We encourage neonatal researchers to report on outcomes in this fashion. Because parents generally report that function is more important than diagnoses, incorporating functional measures for children born preterm is an ethical imperative.

The strengths of this study include leveraging the existing CNFUN infrastructure and follow-up visits to recruit study participants from 13 sites across Canada. We included more than 1000 parents to compare their perception with CNFUN. Second, we had detailed information from the standardized health and neurodevelopmental evaluation on family demographic and clinical characteristics that allowed for investigation of our secondary objective on the association between the discrepancies in parental and CNFUN classifications. Lastly, parents of children born preterm were active members of our research team and involved in every stage of the study from proposal development to data interpretation.

This study also has some limitations. First, our study captures only parental perspectives at the 18- to 21-month CA clinic visit. Parental views and concerns may change at different ages and stages of their child's development. Second, parents were only asked one question about their classification and were not invited to elaborate. This allowed us to capture the perspectives of a large number of participants in scope but limited the depth and nuances of their responses. On the other hand, in other studies by our research group, many parents reported that they value function over diagnosis.[22–24] Third, not all CNFUN follow-up clinics participated, so parent participants from larger, more academic regions may be overrepresented. Three sites were excluded due to logistical issues with recruitment. Fourth, the measure used by parents to rate their child's development was not tested for validity or reliability, but it was created with parental input and used to gather many responses widely across the network by removing barriers to participation and maximizing the number of consenting parents. Lastly, Canada has a universal health care system, a generous parental leave policy, and well-funded daycare in certain provinces. It is unknown whether parents in countries who do not share these characteristics would answer similarly. However, our study with more than 1000 parents' voices suggests that current classifications of NDI used in neonatal follow-up research need to be revisited. Our research group has identified valid questionnaires and tools to measure parent-identified outcomes that reflect child functioning.[44] We are also moving away from labels to reduce the risk of miscommunication when using terms like severe NDI within this preterm population. Future research should examine the acceptability, appropriateness, and feasibility of implementing valid and reliable tools into neonatal follow-up and how to communicate prognostic outcomes with the goal to better align with parental perspectives and values.

CONCLUSION

Parental perception of neurodevelopment in children born extremely preterm differ from the medical classifications used by CNFUN and other networks. In particular, the term

"severe NDI" may be misunderstood. Neonatal follow-up research should consider choosing outcomes that describe functional abilities rather than more subjective categories of mild, moderate, and severe. Collaborative efforts of parents and clinicians/researchers together can improve communication that is vital in clinical practice, health care delivery, and health outcomes for children born preterm and their families.

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ABBREVIATIONS

Bayley-III: Bayley Scales of Infant and Toddler Development, Third Edition
BPD: bronchopulmonary dysplasia
CA: corrected age
CNFUN: Canadian Neonatal Follow-Up Network
CP: cerebral palsy
GA: gestational age
GMFCS: Gross Motor Function Classification System
NDI: neurodevelopmental impairment
NICU: neonatal intensive care unit.

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