



“Quality of life”: parent and neonatologist perspectives

Shannon Y. Adams¹ · Richard Tucker² · Melissa A. Clark³ · Beatrice E. Lechner^{1,2}

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Abstract

Objective To assess how physicians and families understand quality of life (QOL) for NICU patients, and to explore the feasibility of developing a standardized definition for QOL.

Study design Surveys were developed and administered to neonatologists and eligible families. Quantitative analysis was conducted using standard statistical methods. Qualitative analysis was conducted using NVivo software. Focus groups were conducted with the same groups, and audio recordings were obtained and analyzed for recurring themes.

Results Both parents and physicians value QOL as a metric for guiding care in the NICU. Parents were more likely to accept higher levels of disability, while neonatologists were more likely to accept higher levels of dependence on medical equipment. In relation to infant QOL, predominant themes expressed in the parent focus groups were stress levels in the NICU, advocating as parents, and the way in which long-term outcomes were presented by the medical team; in the physician focus group, the ambiguity of predicting outcomes and thus QOL was the main theme.

Conclusions Both parents and physicians recognize the importance of QOL in the decision-making process for critically ill infants, but the two groups differ in their assessment of what QOL means in this context. These data suggest that QOL cannot be adequately defined for standardized use in a clinical context, and as such, should be used thoughtfully by neonatologists in discussions of end- of-life care.

Introduction

Neonatal mortality is defined as death in the first 28 days of life [1]. Roughly two-thirds of all infant deaths before the age of one occur during this neonatal period [2]; in 2017, the Centers for Disease Control reported a neonatal mortality rate of 3.85 per 1000 live births in the United States [1]. Many of these deaths, which can be attributed to a multitude of factors, including prematurity, congenital malformations, and genetic conditions [3], occur in the neonatal intensive care unit (NICU). Numerous studies have explored the emotional and psychological trauma for families who spend time in the NICU [4–6]. It is not uncommon for parents of hospitalized NICU infants to experience post-traumatic stress disorder (PTSD) [7], and

grief is further exacerbated when a child dies in this setting. A study from the University of Michigan focusing on perinatal loss up to 28 days found that 9 months after loss, women showed “remarkably high and persistent levels of distress” consistent with severe depression and PTSD [8].

Neonatal deaths in the NICU setting can be broadly divided into two primary categories, namely (a) “unavoidable,” those with unstable physiology who die despite intensive care or intervention due to unmanageable causes, and (b) “negotiated,” those infants with stable physiology who, in the absence of active anatomical failure, die due to the discontinuation of clinical intervention, such as cardiopulmonary resuscitation and use of a ventilator [9]. The latter often takes prognosis into consideration, as well as what the medical profession calls “quality of life” (QOL). In broad terms, this refers to the general well-being of an individual defined by both negative and positive outcomes [10–12].

The concept of “QOL” is frequently used in clinical discourse; however, from setting to setting, it has no conclusive definition [10]. In fact, QOL can refer to two distinct, if overlapping, entities. One is a broad conception of what QOL means, while the other is health-related QOL, which is more often referenced in the medical literature. The medical field has been trying to measure QOL for decades.

✉ Shannon Y. Adams
shannon_adams@brown.edu

¹ Warren Alpert Medical School of Brown University, Providence, RI, USA

² Women & Infants Hospital of Rhode Island, Providence, RI, USA

³ Brown School of Public Health, Providence, RI, USA

A 1996 landmark study in the *New England Journal of Medicine* conceptualized QOL in terms of multidimensional domains, including social, physical, and psychological, as understood through individual experiences, perceptions, and beliefs [13]. Assessment tools specific to pediatric medicine came later, most notably with the Pediatric Quality of Life Inventory (PedsQL), which has been validated as a means of reliably quantifying health-related QOL outcomes for children with chronic health conditions [14–16]. More recently, the PedsQL Infant Scales were developed, which have also been found to be a valid and reliable means of assessing healthy and ill infants aged 1–12 and 13–24 months [17, 18]. However, even the PedsQL Infant Scales deal only with current health status and expected newborn milestones, as opposed to conceptions of QOL that include long-term medical, psychological, and social/emotional sequelae, or prognostic outcomes based on diagnoses made during the neonatal period.

It can be argued then that a gap in the data exists, particularly as it relates to the explicit delineation of factors that are and should be incorporated into otherwise subjective clinical evaluations of the overall QOL of neonatal patients. This is especially important when considering how “QOL” as an idea is utilized in discussions between physicians and families when determining the best course of treatment for critically ill infants, including those transitioning to comfort or palliative care [19, 20]. Some within the field contend that the fluidity of the term QOL as it is currently employed is beneficial, allowing physicians to evaluate each patient and make informed decisions on a case-by-case basis without the rigidity of guidelines that may stifle their clinical judgment. However, there are others who argue in favor of a more uniform palliative care transition process for neonates, which includes discussions of QOL. Proponents of standardization assert that set policies around these ideas would help to ensure a minimum level of care for all families, facilitating consistency, and supporting providers through what is often a very difficult clinical encounter [21].

Therefore, when considering the implications of the conclusions drawn with QOL as the principal rationale, as well as the diversity of arguments surrounding the utility of more explicit QOL protocols, the aim of our study was to assess how neonatologists and families describe QOL for NICU patients, and to explore the feasibility of developing a standardized definition for clinical use [11, 22].

Methods

Study structure and logistics

This was a two-part study. The first part consisted of an electronic survey to two study groups: neonatologists and

parents or caregivers who have experienced a child in the NICU. Survey development was aided by feedback from family members of former NICU patients and members of the Women & Infants Hospital NICU Family Advisory Council.

NICU families were identified in two ways. First, Internet searches were conducted that generally identified active support or advocacy groups that specifically targeted NICU families. “Family” for the purpose of this survey was defined as “any individual who identified as a parent, guardian, or primary caregiver of a child who spent some time in the NICU”. Groups were defined as “active” if they had one or more posting online within the previous 3 months. The Premie Parent Alliance, now the NICU Parent Network, was one of most significant contributors, distributing the survey via their multiple social media channels to the 37 member organizations under their umbrella.

Next, Internet searches were conducted that specifically identified support or advocacy groups that were tailored to underrepresented persons, including minorities and fathers. Once approached and recruited, all participating groups were asked to distribute the survey through their channel of choice, including but not limited to social media, email, and community newsletters. Additional organizations and individuals not initially contacted were welcomed and encouraged to repost survey links and descriptions obtained from the partnering organizations, although these transactions were not specifically tracked. Data collection for the family survey occurred over a 2-month period.

Surveys of neonatologists were administered to neonatologists in academic divisions through the neonatal–perinatal medicine division chief listserv. Data collection for the neonatologist survey occurred over a 1-month period.

Participants in both surveys were asked to rate the importance of QOL as a metric for critically ill children in the NICU setting. They were then asked to select choices from a predetermined list of 20 disabilities, developed based on common long-term impairments and health outcomes for NICU patients, which they felt were compatible with a “good” or “bad” QOL. Participants were also given the option to provide additional feedback regarding other QOL factors not listed (Table 1). Demographic information was also collected from both study groups.

The second part of the study consisted of focus groups geared toward the collection of in-depth qualitative data on parents’ and neonatologists’ perceptions on QOL for NICU infants. Three focus groups were conducted, two with families and one with neonatologists. The physician focus group was conducted in Providence, RI. The family focus groups were conducted in Baltimore, MD and Providence, RI. Participants for each of the family focus groups were recruited through private NICU support groups in the area. All focus groups were recorded, and the participants signed consent forms prior to the discussion. A Starbucks

Table 1 Parent and neonatologist QOL survey questions.

Question	Subsections	Type of response solicited
1. (a) If you were asked to come up with a definition of BAD quality of life for a child, which items below do you think should be included in the definition? In other words, please pick up to five items that you think are most important in defining BAD quality of life for a child.	Physical disability or appearance: 1. Absent or handicapped limbs(s) 2. Unable to get out of bed/sit up 3. Able to sit but unable to stand or walk 4. Need for orthopedic support while walking, such as braces, walkers, or splints 5. Physically looking different than other children, i.e., disfiguration/dysmorphia Sensory disability: 6. Deafness 7. Hearing impairment but not complete deafness 8. Blindness 9. Need for glasses or contacts Cognitive disability: 10. Inability to obtain a formal education, i.e., intellectual disability/cognitive deficit 11. Nonverbal 12. Learning disorder diagnosis such as ADHD or dyslexia 13. Incontinence	Multiple choice, up to five possible, minimum of required
1. (b) For the next question, please choose the disabilities that you think a child could have and still have a good quality of life. You may choose as many or as few options as you want.	Emotional disability: 14. Inability to interact with others in a formal (e.g., school) or informal (e.g., public locations such as a mall) social settings 15. Mental health problems such as anxiety, depression, schizophrenia, and bipolar disorder. Need for medical care or technologies: 16. Need for one or more future surgeries 17. Need for organ or bone marrow transplant 18. Need for frequent hospital stays 19. Need for a ventilator 20. Need for a tracheostomy or g-tube	Multiple choice, no limit, must choose a minimum of one required
2. What factors do you believe are missing from the list above, which would make for a bad quality of life for a child?	N/A	Additional feedback, not required
3. How important do you think quality of life is in determining whether to continue life-sustaining therapies for infants in the NICU?	N/A	Sliding Likert scale: 1 = not at all important to 10 = very important

\$10.00 gift card was given to participants at the end of each session as a token of appreciation for their participation. The family focus groups included dinner. All groups were scheduled and ran for 1–1.5 h in length.

Data analysis

Comparisons between family and physician survey responses were made using the chi-square test for categorical variables and the Wilcoxon rank-sum test for continuous variables. For variables with cell sizes less than five, the Fisher’s Exact test was used.

All open-ended entries in the survey were analyzed with NVivo software, which was used to visualize the qualitative information in a quantitative fashion, grouping the text based on subject matter, assessing for frequency of word use, and evaluating for similarities between respondents’ entries.

Focus group recordings were manually transcribed and analyzed by the research team looking for thematic patterns, including frequency, intensity, specificity, participant perception of importance, and extensiveness [23]. Theme development was directed by the content of the data. Theme notes from transcribed scripts were analyzed by two members of the research team. Recurrent patterns in meaning were derived from the data, and the most prevalent themes are presented.

Results

Survey

Overall, 112 neonatologists responded to the physician survey. For the family survey, a total of 62 organizations

Table 2 Participating support groups, family survey distribution.

Organization	Location/headquarters	Method of distribution
Hand to Hold	Austin, Texas	Facebook page, Twitter
Project Sweet Peas	Warwick, RI	Facebook page, Twitter
Trisomy 18 Foundation	Dale City, VA	Private Facebook group
Minnesota Premies	Minnesota	Facebook page
Team Grayson	Boston, MA	Facebook page/private group, Twitter
Wills Way Foundation	Chicago, IL	Facebook page
NICU Families NW	Portland and Vancouver, WA region	Private Facebook group
Keep Em' Cookin'		Facebook page, Twitter
European Foundation for the Care of Newborn Infants	Munich, Germany	Facebook page, Twitter
NIDCAP	Boston, MA	Facebook page
Ronald McDonald House of Durham	Durham, NC	Private Facebook group
Hope for HIE (Hypoxia Ischemic Encephalopathy)	West Bloomfield Township, MI	Facebook page
For the Love of Babies	Ventura, CA	Facebook page
National Perinatal Association	Lonedell, MO	Facebook page
Insta Fathers		Email listserv, private Facebook group
National Coalition for Infant Health	Washington D.C.	Facebook Page, Twitter
Preemie Parent Alliance ^a	Madison, MN	Email listserv (including three reminders), public website link, newsletter, and Twitter

^aThe Preemie Parent Alliance is a network of 37 organizations that engage in related advocacy and support for children and families. These groups include Bryce's NICU Project, Connected Forever, Courageous Steps, Families Blossoming, Go Premies!, Graham's Foundation, Hand to Hold, Holding Tiny Hands, Instituto Pequenos Grandes Guerreiros, It's a Preemie Thing, Keep Em' Cookin', Lily's Hope Foundation, Little Giraffe Foundation, Me Two Books, National Perinatal Association, NEC Society, NICU Families NW, NICU Helping Hands, Nurtured by Design, Once Upon a Preemie, Pebbles of Hope, Preemie: Lessons in Love, Life and Motherhood, Preemie World, Project Preemie, Silvie Bells, Speaking for Moms & Babies Inc., Team Grayson, The Family Support Network of Central Carolina, The Gift of Life, The Morgan Leary Vaughan Fund, The PPRM Foundation, The Tangerine Owl Project, the Tiny Miracles Foundation, Triple Heart Foundation, Twin to Twin, Will's Way Foundation, and the Zoe Rose Memorial Foundation.

both domestically and internationally were contacted via email, Facebook, and telephone; 45 agreed to participate, including the 37 organizations within the Preemie Parent Alliance Network (Table 2). All participating groups distributed the survey through their channel of choice, including Facebook page, Facebook group, email, Twitter, public website, and community newsletters. In total, 901 family member responses were recorded.

Table 3 describes the survey study population across both groups.

Quality of life: disability ratings

Both groups were asked to rate the importance of using QOL as a metric when determining whether to continue life-sustaining therapies for infants in the NICU. There was no difference between the groups in the median rating of the

importance score. On a scale of 1–10, physicians rated the importance 8.1 (SD 15.6) and families 8.2 (SD 27.2).

Respondent selections of disabilities associated with “bad” and “good” QOL were tabulated and ranked according to the frequency of selection. Table 4 shows the percentage of respondents who selected each disability associated with a “bad” QOL. A ranking of 1 indicates that the greatest number of parents or physicians believed that the given disability is associated with a “bad” QOL for a NICU child. Physicians were more likely than families to associate inability to interact with others, intellectual disability, inability to speak, inability to get out of bed, absent or severely handicapped limbs, and deafness and incontinence with a “bad” QOL. Families were more likely than physicians to associate the need for a ventilator, need for a tracheostomy/g-tube, and need for future surgery with a “bad” QOL.

Table 3 Demographic data comparisons: families and neonatologists.

Characteristic	Families, n = 901 n (%)	Physicians, n = 112 n (%)
Female	867 (96.2)	57 (50.9)
Age		
Under 30	193 (21.4)	1 (0.9)
30–50	678 (75.2)	57 (50.9)
50+	24 (2.6)	52 (46.4)
Race		
Asian	11 (1.2)	11 (9.8)
Black/African-American	9 (1.0)	5 (4.5)
Caucasian	794 (88.1)	80 (71.4)
Hispanic	42 (4.7)	9 (8.0)
Native American	8 (0.9)	0 (0.0)
Pacific Islander	3 (0.3)	1 (0.9)
Other	26 (2.9)	0 (0.0)
Marital status		
Married/living as married	799 (88.7)	98 (87.5)
Widowed	1 (0.1)	0 (0.0)
Separated/divorced	42 (4.7)	4 (3.6)
Single/never married	50 (5.5)	10 (8.9)
Religious attendance		
Weekly	158 (17.5)	24 (21.4)
Not weekly but more than just holidays	194 (21.5)	22 (19.6)
Only holidays	68 (7.5)	17 (15.2)
Almost never	244 (27.1)	21 (18.6)
Never	208 (23.1)	23 (20.5)
Region of residence		
New England	90 (10.0)	20 (17.9)
Mid-Atlantic	101 (11.2)	22 (19.6)
Midwest	163 (18.1)	11 (9.8)
Southeast	86 (9.5)	38 (33.9)
South-Central	96 (10.7)	12 (10.7)
Mountain	56 (6.2)	1 (0.9)
Pacific	87 (9.7)	4 (3.6)
Children		
Are a parent	N/A	95 (84.8)
Have a child w/disabilities/medical issue	375 (41.6)	6 (5.4)
Have a child who died in the NICU	11 (1.2)	0 (0.0)
Highest level of education		
Some HS	6 (0.7)	–
HS degree	54 (6.0)	–
Vocational/Tech.	41 (4.6)	–
Some college	126 (14.0)	–
Assoc. degree	91 (10.1)	–
Bach. degree	341 (37.8)	–
Grad. degree	232 (25.7)	–

Table 3 (continued)

Characteristic	Families, n = 901 n (%)	Physicians, n = 112 n (%)
NICU length of stay		–
Less than 7 days	95 (10.5)	–
7–28 days	293 (32.5)	–
29 days to less than 3 months	349 (38.7)	–
More than 3 months	273 (30.3)	–
No. of children in the NICU		–
0	2 (0.2)	–
1	647 (71.8)	–
2	205 (22.8)	–
3	41 (4.6)	–
4	4 (0.4)	–
5	2 (0.2)	–
Type of practice		
Academic	–	106 (94.6)
Private	–	4 (3.6)
Other	–	2 (1.8)

Table 5 shows the percentage of respondents who selected each disability when considering which of the options allow for a “good” QOL. A ranking of 1 here indicates that the highest percentage of respondents believed that the given disability does not preclude a child from having a “good” QOL. Parents were more likely than physicians to allow the definition to include absent or handicapped arms or legs, inability to get out of bed, blindness, intellectual disability, inability to speak, inability to interact with others, and mental health problems. Physicians were more likely than parents to tolerate a need for future surgery and need for a tracheostomy.

Respondents were able to select any number of disabilities they believed an infant could have and still have a “good” QOL. The mean number of disabilities selected by each group was calculated. Family respondents selected a mean of 12.6 disabilities, while physicians selected 11.7 as being compatible with a “good” QOL ($p = 0.087$).

Additional feedback

A total of 281 parents (31.2%) and 52 physicians (46.0%) provided additional feedback when asked “What factors do you believe are missing from the list above, which would make for a bad QOL for a child?” In the family group, the most frequently used term was “pain” with 59 individual references, followed by “vegetative” (25). Lack of “family” (21), lack of “love” (20), and the inability to “communicate” (17) was also discussed as contributory to a “bad” QOL. The majority of respondents also wrote about lack of capacity for joy, the

Table 4 “Bad” QOL rankings.

“Bad” QOL disabilities	Families, <i>n</i> = 901		Physicians, <i>n</i> = 112		<i>p</i> *
	Overall ranking	No. who selected <i>n</i> (%)	Overall ranking	No. who selected <i>n</i> (%)	
Need for a ventilator	1	576 (63.9)	5	44 (39.3)	<0.001
Unable to get out of bed/sit up	2	513 (56.9)	1	79 (70.5)	0.008
Need for a tracheostomy or g-tube	3	316 (35.1)	11/12	12 (10.7)	<0.001
Nonverbal	4	235 (26.1)	2	74 (66.1)	<0.001
Incontinence	5	232 (25.7)	6	39 (34.8)	0.047
Inability to obtain a formal education/intellectual disability	6	223 (24.8)	4	54 (48.2)	<0.001
Inability to interact with others in a formal or informal social setting	7	197 (21.8)	3	56 (50.0)	<0.001
Absent or handicapped limbs(s)	8	194 (21.5)	7/8	34 (30.4)	0.040
Need for frequent hospital stays	9	177 (19.6)	9/10	15 (13.4)	
Need for organ or bone marrow transplant	10	113 (12.5)	9/10	15 (13.4)	
Able to sit but unable to stand or walk	11	95 (10.54)	11/12	12 (10.7)	
Blindness	12	83 (9.2)	7/8	34 (30.4)	<0.001
Mental health problems	13	81 (9.0)	14	10 (9.0)	
Need for one or more future surgeries	14/15	65 (7.2)	16/17	2 (1.8)	
Physically looking different than others	14/15	65 (7.2)	15	6 (5.4)	
Deafness	16	46 (5.1)	13	11 (9.8)	0.044
Need for orthopedic support to walk	17	18 (2.0)	16/17	2 (1.8)	
Learning disorder diagnosis	18	12 (1.3)	18/19/20	0 (0.0)	
Hearing impairment, not deafness	19	10 (1.1)	18/19/20	0 (0.0)	
Need for glasses or contacts	20	2 (0.2)	18/19/20	0 (0.0)	

*Unlisted *p* values were not statistically significant.

experience of uncontrollable pain, and the lack of community resources and support as contributing to a “bad” QOL. Some family members (34) also expressed that nothing, including any of the listed outcomes, inherently yields a bad QOL for a child.

In the physician group, the most frequently used terms when responding to the question about “bad” QOL were “inability” (17), referring predominantly to the attainment of developmental milestones, lack of “family” (11), lack of meaningful “interact[ion]” (10), “pain” (7), and “cognitive” impairment or delay (6). Similar to the familial group, physician respondents wrote about the importance of support networks for both the child and the caregiver, as well as the value of connection with loved ones. However, the majority of respondents chose to focus on the relevance of physical disability and intellectual impairment, with zero physicians expressing the belief that a “bad” QOL based on long-term health outcomes does not exist.

Focus groups

The family focus groups included seven and four participants. In these groups, QOL was generally divided into two categories: in the NICU and long term. QOL in the NICU focused primarily on two areas. The first was the stress level that intensive care interventions place on infants, as expressed by the following quotes:

“There was just so much stress – the beeps, the pricks the noise. It was too much.”

“I can’t just do normal mom stuff. When my baby is crying and I can’t just pick her up...you have to ask permission, there are wires, there are all sorts of things, it’s weird.”

Table 5 “Good” QOL rankings.

“Good” QOL disabilities	Families, <i>n</i> = 901		Physicians, <i>n</i> = 112		<i>p</i> *
	Overall ranking	No. who selected <i>n</i> (%)	Overall ranking	No. who selected <i>n</i> (%)	
Need for glasses or contacts	1	835 (92.7)	2	103 (92.0)	
Hearing impairment, not deafness	2	799 (88.7)	1	105 (93.6)	
Need for orthopedic support to walk	3	783 (86.9)	3	98 (87.5)	
Learning disorder diagnosis	4	747 (82.9)	5	92 (82.1)	
Deafness	5	710 (78.8)	7	76 (67.9)	0.006
Physically looking different than others	6	672 (74.6)	6	95 (84.8)	
Need for one or more future surgeries	7	659 (73.1)	4	96 (85.7)	0.007
Blindness	8	652 (72.4)	11/12/13	63 (56.3)	<0.001
Absent or handicapped limbs(s)	9	622 (69.0)	14	61 (54.5)	0.001
Mental health problems	10	597 (66.3)	11/12/13	63 (56.3)	0.027
Able to sit but unable to stand or walk	11	555 (61.6)	20	63 (56.3)	
Need for organ or bone marrow transplant	12	551 (61.2)	8	69 (61.6)	
Need for frequent hospital stays	13	477 (52.9)	9	67 (59.8)	
Incontinence	14	470 (52.3)	15	53 (47.3)	
Inability to obtain a formal education/intellectual disability	15	456 (50.6)	17	41 (36.6)	0.004
Nonverbal	16	435 (48.3)	18	31 (27.7)	<0.001
Need for a tracheostomy or g-tube	17	400 (44.4)	10	66 (59.0)	0.005
Inability to interact with others in a formal or informal social setting	18	398 (44.2)	19	28 (25.0)	<0.001
Need for a ventilator	19	271 (30.1)	16	42 (37.5)	
Unable to get out of bed/sit up	20	239 (26.5)	11/12/13	17 (15.2)	0.008

*Unlisted *p* values were not statistically significant.

The second area was the need for parents to advocate for their child’s well-being during their hospitalization. Many parents noted that they believed themselves to be the expert on their child, and therefore found it important to be seen as an equal when making decisions alongside the medical team.

“You’re just so numb. You’re tired, you just had a baby, you’re having so much information thrown at you so fast that you have no choice but to trust them. But you have to speak up; you have to find your voice.”

“As I got more comfortable and as I learned the terminology and as I learned my baby, I was able to help make care decisions for her. And I think that was huge because eventually towards the end, I told the doctors that we were pulling her off of oxygen because I knew she could handle it...so I think it was

good that I was seen as a part of the care team and that they listened to me.”

Regarding QOL long term, family participants spoke mostly about the difficulty around, and personal resistance to, conversations of long-term QOL in the NICU, contrasted by the desire to be thoroughly informed by the medical team. The also stressed the important of not being pressured to think of severe impairment as an inherently negative outcome, particularly when considering the power that physician advice has on the decisions families ultimately make. For all participants, the way that discussions about QOL and long-term outcomes for the child were handled by the medical team played a large role in their perceptions of treatment and satisfaction with care. The representative quotes included:

“You’re still trying to get through that day, never mind when they are seven or eight...to wrap your

head around quality of life when your child is an infant is impossible.”

“The medical team cannot possibly know. They can give you statistics and research, but with a fragile infant it really could be anything.”

“It really was helpful for me when talking about prognosis and quality of life, when my doctors would use the clinical terms but then they would take the time to help me understand what they mean in real terms. Because I don’t have a medical degree, and to hear it in a way that I could understand and what the implications for my daughter were going to be, that was everything.”

“Everybody was constantly warning me. My son has a lot of those [disabilities] that were named and for me none of those things matter. He can’t talk, can’t walk – but he can be happy and sad, and he knows me, he knows dad, he knows his siblings, he feels joy and he laughs and he smiles. He can feel all those things, so for me that is quality of life.”

The physician focus group included five participants. This group discussed the levels of ambiguity in predicting long-term outcomes for NICU patients. But despite this, all of the participants felt strongly that a theoretical line exists where care should be redirected from interventional to comfort based. In addition, though all of the neonatologists present found irreplaceable value in teamwork, including the voice and wishes of the parent, they also acknowledged the role of the physician as the leader in these discussions. The representative quotes include:

“There are babies with such profound impairment that they are not able to interact with their environment, experience joy, what makes us human. I think most people would accept that threshold. Self-awareness, inability to experience love and reciprocate that, then that’s an outcome worse than death.”

“There are very few instances of people trying to move to redirect care too early. I think the problem more often is the lack of recognition soon enough that it is time to change the course of care.”

“Advocates for parental autonomy are usually not clinicians. They call it paternalistic. But it is an

incredibly difficult thing for a mother to say, ‘I want you to stop.’ Many people are looking to you as the clinical leader to say, “This is what I recommend.” They want you to take that burden off of them.”

Discussion

Before interpreting the results, it is important to understand the reason that the two study groups were chosen, and to do that we must first define family-centered care (FCC) and the process of shared decision-making in the NICU. As one study states, FCC is “an approach to medical care rooted in the belief that optimal health outcomes are achieved when patients’ family members play an active role in providing emotional, social, and developmental support” alongside the physician [24]. The cornerstone of FCC in the NICU is the belief that families should actively participate in the determination of their child’s care, thereby facilitating physician and parent collaboration [25]. As this is the philosophy in most NICUs today, both parent and physician perspectives must be explored in order to thoroughly examine interpretations of QOL and its utility as a clinical metric.

With this in mind, we can better contextualize the significance of data collected. The findings reported here indicate several key differences between how families and physicians evaluate QOL in the NICU setting, when focusing on the specific disabilities analyzed. One example of these differences is the perception of medical intervention. Families were more likely than physicians to associate the need for a ventilator, need for a tracheostomy, and need for future surgery with a “bad” QOL, while physicians were more likely than parents to allow the definition of a “good QOL” to include need for future surgery and need for a tracheostomy. Parents also expressed concern regarding the NICU environment itself, identifying the setting as “stressful,” and referencing the continuous “poking and prodding” as a perceived threat to the patients’ ability to develop naturally. Physicians, however, did not raise this concern. This incongruity between parent and staff perceptions of infant discomfort due to medical intervention was likewise identified in a 2004 study in California by Gale et al. They found that NICU parents felt as though the team often minimized their child’s pain when enduring routine procedures, and expressed a desire for the staff to see and respond to the infants’ pain as a parent would [26]. Similar studies, including a 1997 paper by Wereszczak et al., also observed parental distress by the perceived pain that infants sustained during standard clinical practice in the NICU [27, 28]. This discrepancy in perception between parents and physicians can most likely be attributed to

familiarity and understanding. While parents may see one child with a tracheostomy, neonatologists will treat many over the course of their careers, particularly in high-acuity centers [29]. Furthermore, years of clinical practice allow physicians to better understand the utility of these technologies in patient care than parents. As one physician in the focus group said, "We do things to babies that are uncomfortable, painful, stressful, and it's acceptable to make those interventions to cause that pain and discomfort because it serves a purpose of survival".

Another key divergence was found in the interpretations of long-term health outcomes. In general, physicians were more concerned than families with enduring physical, mental, and cognitive impairment of children. For example, thirty-four parents expressed in their written comments a belief that no combination of the listed disabilities constituted a bad QOL for a child, while no physicians expressed this view. There are multiple ways to interpret this finding. First, as Steinhäuser et al. demonstrated, it may reflect a difference in prioritization of QOL factors between physicians and patients/parents [30]. This difference may stem from what some activists and bioethicists refer to as "medical ableism," or the "assumption that the 'normal' able body is better than abnormal bodily forms" [31]. According to some, medicine as a profession has historically underestimated and undervalued the QOL of persons with disabilities [32]. Many studies have shown that what physicians perceive to be detrimental to QOL, patients with disabilities do not [33, 34]. In the setting of the NICU, our study shows that parents likewise believe that extensive disabilities do not preclude a good QOL for babies, reflecting positive parental associations with raising a disabled child, as numerous studies have shown [35–37]. As one respondent wrote in the additional feedback portion of the survey, "None of these are reasons to think your child would have a bad QOL. Challenges? Sure—but they could still have a happy life".

Another possibility may be a lack of experience on the part of parents when compared to the training and expertise of physicians. As many in the focus groups mentioned, parents in the NICU are often encountering these situations for the first time. Most have no formal education in the science underlying the care of infants, and the intense nature and sheer stress of the NICU setting may further prevent families from being able to rationalize the challenges ahead [4, 38, 39]. Neonatologists, on the other hand, will have treated similar patients and seen the various sequelae of similar diagnoses, and thus view outcomes from a more data-driven, rational perspective. As one physician in the focus group said, "Oftentimes you know when something will not turn out well for the family. Many of us have been doing this for a long time and these are conversations we are very comfortable and well-versed in". However, it is

important to note that exposure to and practice treating medically complex patients in the NICU does not equate to the experience parents have raising these children at home. In total, 41% of the participants in our family survey were identified as caretakers of children with medical problems, and although they may not have the same knowledge base to understand the ramifications of certain diagnoses as their child's physician, they do have a different kind of knowledge about what a patient's life looks like after they leave the hospital. Furthermore, clinical decision-making is an imperfect science. A study by Blanco et al. [40] found that the medical profession routinely underestimates survival rates and overestimates long-term disability rates for premature infants, particularly at less than 28 and 26 weeks of gestation [40]. Therefore, while neonatologists' level of training may allow them to guide a family to see their infant beyond a snapshot in time, it may not always be true that a QOL projection made by a neonatologist based on prognostic indicators is inherently more accurate than that patient's family.

A third explanation reflects the relational connection between parent and child. As both groups mentioned, it is extremely difficult to have a conversation about transitioning an infant to comfort care, especially when that child is your own. In the focus groups, several participants expressed a willingness and even desperation to go home with their baby alive, even if that meant adjusting to "a new normal", namely disability or morbidity. This becomes especially true for families who spend long periods of time in the NICU. As one mother said, "Your perception of QOL changes with each day in the hospital. After a while you just want to get started on your life—you don't care what it looks like". The emotional bond parents feel, as well as perceived parental responsibility to protect, may cloud their ability to rationally weigh difficult decisions. One study addressed the concept of "good parenting" for sick children, and noted that many parents believed that asking for more treatment or more time, whatever the cost, is synonymous with being a better parent [41]. Although physicians often feel deep connections with patients and their families, the relationship with a patient is based on a different set of goals. As one doctor said, "We are primarily driven by what's in the best interest of the baby. Often the family may not be there yet—still resisting the inevitable. We are there to help them educate themselves, use the information available to make a decision that limits pain and suffering whenever possible".

It is also important to highlight the similarities between the two groups studied. Both groups found similar value in QOL as a metric to consider when determining whether to continue life-sustaining therapies for infants. This foundational, shared belief is important because it demonstrates the relevance of this discourse and related research. Members of both groups also referenced uncontrollable pain as a barrier

to QOL that warranted transitions to palliative or comfort care. Another similarity was found in discussions of connection and communication with loved ones as a measure for QOL. Both groups referenced the concept frequently in the free response section of the survey, and “inability to interact with others” was ranked low at 18 and 19 out of 20 by parents and physicians respectively as being compatible with a “good QOL”. This reflects a universal recognition of the role that community, support, and nurturing play in the health and well-being of NICU patients, with the power of relationships as a defining feature for QOL.

Our data demonstrate overlap as well as key differences in physician and parent understanding of both good and bad QOL in NICU infants, which renders the development of a clearly defined QOL metric for use as a clinical decision-making tool in the NICU, to be a challenging undertaking. Some experts believe that there is utility in standardizing the way physicians talk about the care of NICU patients in the context of QOL and palliative care transition [42, 43]. Other researchers have acknowledged the role that systemic protocols play in the preservation of distributive justice, referring to a need to strategically allocate resources and therefore choose not to provide futile treatments to those babies for whom there is little chance of survival [44]. However, the trouble as our data demonstrate, is the immense variability in interpretation and valuation of QOL both within and among relevant parties. As one physician in our focus group said, “Is QOL ever so bad that it is worse than non-survival? That gets into philosophical, religious, and other realms where reasonable people can disagree”. Perhaps then, as this study suggests, QOL is unlike ventilatory settings or blood pressure values, and cannot be reliably defined or standardized for use in a clinical context. Since the term QOL bears so many connotations, signifying different concepts depending on one’s role and perspective, it cannot be used in a meaningful way among a team of individuals trying to determine the course of treatment for an infant. Therefore, despite its prevalence in NICU discourse, we conclude that the common practice surrounding how infant QOL is discussed is problematic, and that neonatologists should use QOL judiciously when communicating with families regarding care, knowing that it does not in fact have a validated definition acknowledged by the medical or parent communities.

One area of the larger QOL debate that is not addressed in this study is the idea of quantity versus quality of life. Longevity, and the projected number of years lived, has historically been a metric used by the medical field when determining prognosis and treatment course. The NICU is no different. There has been some research that incorporates both issues, using NICU graduates who have survived to adolescence and adulthood to assess health-related QOL scores. In one such study, children with former diagnoses of

extreme prematurity and very low birth weight reported a lower health-related QOL than their term-born counterparts, with significant difficulties in economic and social function despite survival to early adulthood [45]. Another study, however, conducted with a similar patient population, found that health-related QOL for those who survived to young adulthood was not related to size at birth or the presence of disability [46]. More research is needed to understand the intersection of total life years and experienced QOL, both from the patient and family perspectives.

A limitation to this study is the demographics of the familial participant population, namely that family participants in the survey were primarily white and female. This lack of diversity, despite a targeted recruitment process, limits the generalizability of the findings. Unfortunately, these pitfalls are common to survey and parent-based research in the neonatal population. For one, pediatric research has historically struggled to include the perspective of fathers [47]. One 2005 study focusing on paternal representation in parenting and child psychology research found that only 2.3% of studies in health-related journals included the assessment of fathers only, compared with 46.7% focusing on mothers only [48]. This challenge is pervasive across other pediatric disciplines as well [49]. Achieving racial diversity in research recruitment and participation has historically been problematic as well [50, 51]. Similarly, structured studies focused on the neonatal population have encountered these issues (Shelkowitz et al. [52], Cacciatore et al. [53]), indicating a larger need for the concentrated exploration of narratives belonging to parents and families who identify with underrepresented racial and ethnic groups.

Conclusion

This study shows that both physicians and families recognize the importance of QOL in the decision-making process for critically ill infants, but they differ in their assessment of what QOL means for NICU patients. Though both families and physicians are in agreement regarding futility in cases of uncontrollable pain and brain death, parents are more concerned with invasive interventions and the use of medical technology, while physicians focus more on disability and long-term health outcomes. Thus, our findings indicate that QOL cannot be adequately defined for standardized use in a clinical context, and as such, should be used thoughtfully by neonatologists in discussions of end-of-life care.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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