Journal of Pediatric Nursing 66 (2022) 36–43

Contents lists available at ScienceDirect

Journal of Pediatric Nursing

journal homepage: www.pediatricnursing.org

Caregivers' perception of the role of the socio-environment on their extremely preterm child's well-being

Crisma J. Emmanuel [a,*], Kathy A. Knafl [a], Sharron L. Docherty [b], Eric A. Hodges [a], Janice K. Wereszczak [c], Julie V. Rollins [c], Rebecca C. Fry [d], T. Michael O'Shea [c], Hudson P. Santos Jr [e]

[a] University of North Carolina, School of Nursing, Carrington Hall, Campus Box #7460, Chapel Hill, NC 27599-7460, United States of America
[b] Duke University, School of Nursing, Durham, NC, USA
[c] University of North Carolina, School of Medicine, 101 Manning Drive, Chapel Hill, NC, USA
[d] University of North Carolina, Envir. Sciences and Engineering, Gillings School of Public Health, Chapel Hill, NC, USA
[e] School of Nursing and Health Studies, University of Miami, Coral Gables, FL

ARTICLE INFO

Article history:
Received 10 February 2022
Revised 29 April 2022
Accepted 6 May 2022

Keywords:
Determinants of health
Mother-child relationship
Coping behaviors

ABSTRACT

Purpose: The purpose of this qualitative descriptive study was to explore primary caregivers' perception of how social-environmental characteristics, and their own role as primary caregivers, affected their extremely preterm adolescent's well-being.
Methods: Participants were 20 mothers who identified as the primary caregiver of an adolescent born extremely prematurely (<28 weeks gestation) enrolled in the ELGAN cohort study. Data was collected through individual interviews and was analyzed using inductive content analysis.
Results: A total of three themes, and five subthemes, were identified. The two main themes were "familial impact to health and well-being," and "contributors and barriers at the community level." This study described specific familial and community contributors to child and caregiver well-being, including: the importance of advocacy, participating in community activities, and social and familial support networks.
Conclusions: Overall, while there are individual level characteristics that contribute to well-being, a support structure at the family and community level is essential to children born extremely prematurely, and their mother's, well-being.
Practice implications: Healthcare providers caring for these families should understand that not only are extremely preterm youth affected by prematurity, but caregivers are also deeply impacted. Therefore, it is essential that maternal and family care is emphasized by nurses and healthcare providers.

Introduction

Despite dramatic improvements in mortality, adverse neurodevelopmental and long-term negative health outcomes remain a significant concern for extremely premature (EP) children (born <28 weeks gestation) (Joseph et al., 2016; Kuban et al., 2016; Luu et al., 2017; Serenius, 2016). EP children are at a higher risk for neurodevelopmental and psychiatric disorders, such as cognitive impairment, cerebral palsy, anxiety, and depression (Harding et al., 2019; Kuzniewicz et al., 2014; Mathewson et al., 2017; Perapoch et al., 2021; Taylor & O'Shea, 2022). Additionally, children born EP have overall higher rates of physical comorbidities, such as respiratory and cardiometabolic conditions (Crump et al., 2019; Luu et al., 2017). Yet, recent studies have identified children who are able to stay free of adverse outcomes despite the biological insults associated with EP birth (Bangma et al., 2018; Bangma et al., 2019). Additionally, quality of life for most individuals born EP has been found to be comparable to the general population (Roberts et al., 2013; Saigal, 2016).

Previous research in the EP population has focused on identifying early childhood risk factors for adverse health and well-being (Barnett et al., 2018; Doyle et al., 2015; Janevic et al., 2018). Of the research that does explore protective factors, there has been a primary focus on early childhood development (Evans et al., 2014; Granero-Molina et al., 2019). Most studies have utilized quantitative data to explore general protective factors, such as higher socioeconomic status (SES) (Bangma et al., 2019; Benavente-Fernández et al., 2019), higher maternal education (Joseph et al., 2018), and higher infant gestational age (Bangma et al., 2018; Bangma et al., 2019). However, there is limited

* Corresponding author.
E-mail addresses: emmanc@email.unc.edu (C.J. Emmanuel), kknafl@email.unc.edu (K.A. Knafl), doche002@mc.duke.edu (S.L. Docherty), Eric.A.Hodges@unc.edu (E.A. Hodges), jwereszc@med.unc.edu (J.K. Wereszczak), julie.rollins@unc.edu (J.V. Rollins), rfry@unc.edu (R.C. Fry), moshea52@email.unc.edu (T.M. O'Shea).

https://doi.org/10.1016/j.pedn.2022.05.005
0882-5963/

contextualization of how these socio-environmental factors can affect child health from the caregivers' perspective.

For this paper, we conceptualized well-being as an outcome indicator of resilience. Resilience was conceptualized as a dynamic process that allows an individual or family to cope with adversity, change, and opportunity in a manner that results in optimum individual, and family, functioning and well-being (Richardson, 2002; Rutten et al., 2013; Walsh, 2003). Well-being reflects a person's health and life satisfaction in three domains—physical, mental, and social (Ochiai et al., 2021; Vik & Carlquist, 2018).

Of the research that focuses on protective socio-environmental factors for EP individuals, in-depth family and community dynamics are often difficult to fully describe due to their complexity and researchers' focus on objective measures and outcomes (Morgan et al., 2022; Saigal, 2016). Therefore, we aimed to look at this phenomenon through the perspective of primary caregivers of EP individuals. While traditionally a parent, we defined a primary caregiver as anyone who took primary responsibility for the preterm child's ongoing care (Family Caregiver Alliance, 2021) across the child's lifespan. Currently, several researchers have been able to identify how vital primary caregivers are to an EP child's well-being across childhood, often acting as both the child's primary caretaker and advocate (Evans et al., 2014; Granero-Molina et al., 2019). In addition, caregivers are often deeply affected by the birth of an EP child, often viewing preterm birth as a traumatic and stressful experience (Gonçalves et al., 2020; Holditch-Davis et al., 2015).

Therefore, the aims of this paper were to 1) describe primary caregivers' perception of social-environmental characteristics they believed affected their child's, and their own, well-being, and 2) describe how primary caregivers view their role in fostering their child's well-being.

Methods

Study design

We used a qualitative descriptive design due to its philosophical and methodological foundation (Doyle et al., 2020; Sandelowski, 2010). A qualitative descriptive design allowed us to base our study on the philosophical foundation that reality is multiple and subjective (Doyle et al., 2020; Long et al., 2018). Additionally, the focus for our analysis was to maintain results that stayed close to the participants' own views, and which could be useful to future intervention development and clinical practice (Doyle et al., 2020; Long et al., 2018; Sandelowski, 2000).

Sampling & Setting.

Data came from a larger, mixed method study focused on participants in the Extremely Low Gestational Age Newborn (ELGAN) cohort study. ELGAN is one of the largest longitudinal, cohort studies (data collected at ages 2, 10 and 15 years of age for children born extremely preterm) of Extremely Preterm Children (O'Shea et al., 2009; Taylor & O'Shea, 2022). ELGAN is focused on neurological, positive health, psychiatric and behavioral outcomes (O'Shea et al., 2009).

A purposive sample from the ELGAN cohort study was recruited to achieve maximum variation, specifically variations in neurodevelopmental and racial/ethnic backgrounds (Palinkas et al., 2015; Patton, 1990). We selected this sampling method to both describe variations in experiences and explore core elements and shared outcomes (Patton, 1990). Therefore, our criteria were 1) a primary caregiver of a child currently enrolled in the ELGAN cohort study; 2) identified as the primary caregiver from the child's early childhood to present; 3) able to give informed consent.

This study took place in the southeastern United States (U.S.). Due to COVID restrictions, we were unable to conduct interviews in participants' homes as originally planned. Instead, primary caregivers were given the option of being interviewed via a telephone or Zoom call. While most participants were in the southeastern U.S., a small percentage lived in other areas on the eastern coast.

Recruitment

The primary investigator (PI) conferred with the clinical site coordinator at the University of North Carolina at Chapel Hill's (UNC-CH) ELGAN Cohort study site to identify primary caregivers who met the inclusion criteria and were interested in learning more about the study. The study coordinator was the first point of contact. Primary caregivers who agreed to receive more information provided the study coordinator with a phone number and time for the PI to call to provide additional information. A total of 23 caregivers expressed interest and were contacted by the PI. The PI then offered a verbal description of the study's aims. Caregivers who expressed interest in participating scheduled a follow-up interview visit with the PI and received an emailed written document that provided a detailed description of the study's aims and consent form.

By protocol, if respondents disclosed difficult life circumstances, they would be referred to ELGAN study's clinical coordinator for resources and information and would receive follow up contact with the clinical site coordinator. Additionally, if necessary, the interview would be discontinued. Participants received an overall summary of anonymous results in an annual ELGAN cohort newsletter.

Ethical considerations

This study was approved by the institutional review board (IRB) at UNC-CH. The IRB granted approval to waive signed written informed consent, citing this study as a minimal risk to participants. At the start of the scheduled interview, the PI verbally reviewed the overall aims and consent form that had been emailed. Emphasis was placed on informing participants that there were no penalties for choosing not to participate or to leave the interview at any time. They also were informed of the option of withdrawing their data at any point during the study. Of the 23 that expressed interest, 20 agreed to be interviewed and were compensated with a gift card for their time. No participant elected to stop the interview or withdraw from the study.

Data collection

Semi-structured interviews were conducted via a secure Zoom link or via telephone at a date and time convenient to the participant. An interview guide was developed based on input and feedback from clinicians and researchers familiar with both the ELGAN cohort and family research (see Table 1). In addition, our conceptualization of resilience and well-being informed the development of the interview guide. All the interviews were conducted by the PI, who is a bi-cultural and bilingual (English and Spanish) researcher with extensive training in child development and family health. Based on participants' preferences, 19 interviews were conducted in English, and one was conducted in Spanish using a professionally translated copy of the interview guide. Interviews lasted from 30 min to two hours, and an average of 30–45

Table 1
Semi-structured interview guide.

Questions	Focus
1) Can you describe your child's current health?	General Health
2) Can you describe their Emotional health?	Emotional Health
3) Can you tell me about your child's current wellbeing/Quality of Life?	Well-Being
4) Thinking back on your child's life, can you think of experiences he/she had that were especially important turning points in shaping their health?	Development of Health
5. What about turning points in shaping their well-being/quality of life?	Development of Well-Being
6) Looking back on your child's life, what stands out as especially important in contributing to his/her health	Contributions to General Health
7) What stands out as especially important in contributing to his/her well-being/quality of life?	Contributions to Well-Being

min. 19 interviews were audio-recorded with the participant's consent and transcribed verbatim by the PI. One participant did not want to be audio-recorded but consented to being interviewed—with the interviewer taking detailed notes and verbatim quotes from the participant. All data were de-identified and saved on an encrypted drive. Audio recordings were deleted after transcripts were validated for accuracy. Accuracy of transcripts were validated by the PI listening to taped interviews twice and adding corrections/additions to transcripts as needed. The PI kept a field journal throughout data collection to preserve analytic insights. Field notes were reviewed and used during analysis to further analyze interview data.

Data analysis

Dedoose (v9.0.7, SocioCultural Research Consultants, LLC, Los Angeles, CA), a qualitative data analysis software program, was utilized to assist with data organization and analysis. Inductive content analysis was conducted (Elo & Kyngäs, 2008). The PI read and re-read all transcriptions to gain familiarity with the data set. The PI then completed initial coding of transcripts and field notes. Codes were defined as segments of text based on similar meaning or words (Elo & Kyngäs, 2008). A codebook was then developed based on caregivers' own words and iteratively refined by the PI under the mentorship of a second researcher (HS). Codes were then iteratively organized into categories, which were defined as codes with similar meaning but with a higher level of abstraction than codes. The study's research aims, and the National Institute on Minority Health and Health Disparities Research (NIMHD) Framework, were then utilized to further analyze and organize categories into final subthemes and overarching themes. This methodology is based on prior literature stating that even while utilizing inductive content analysis, the study's aims, and prior theoretical models, can still contribute to data analysis (Armat et al., 2018; Elo & Kyngäs, 2008). Rigor was kept by having the PI maintain an audit trail of memos and PI debriefing with HS. Additionally, the PI was blinded to diagnoses until after data was analyzed. Finally, the PI and HS reviewed the subthemes and themes to come to a final consensus (Morse, 2015).

Code saturation was observed after one fourth of the interviews were reviewed; however, meaning saturation of general experiences was only achieved after interviewing all participants (Hennink et al., 2016). While we included children with a range of neurodevelopmental and health outcomes, meaning saturation only related to the general experience of mothers raising an extremely premature child. Therefore, we were unable to compare experiences by variations in health and/or diagnoses.

Results

In total, 20 primary caregivers, all of whom identified as the mother, agreed to participate in the study. The age of the mothers' child(ren) varied from 16 to 18 years of age at the time of data collection. In addition, racial backgrounds, as well as number and type of comorbidities of their EP child, were varied, but generally reflected the demographic data of the larger ELGAN cohort study. In total, 5% of mothers identified as Latinx, 35% as African American/Black and 60% as White. Of the mothers' preterm child(ren), 5% had no diagnoses, 48% had a physical diagnosis and 62% had a developmental disorder/condition (see Table 2). Of those children who had a physical diagnosis, only 5% of the sample described impairments in activities of daily living.

Overall, there were two major themes identified: 1) Familial Impact to Health & Well-being and 2) Contributors & Barriers at the Community level.

Theme I. Familial impact to health & well-being

The first theme focused on environmental and social factors at the family level and consisted of three subthemes: a) *The Central Role of Motherhood*, b) *The Child's Contribution to Health and Well-being*, and c) *The Family Unit*.

The central role of motherhood

Throughout each interview, mothers emphasized the central role they played in their EP child's ongoing development and well-being. Of the 20 mothers interviewed, all identified themselves as the primary caregiver throughout their child's life. For mothers who described inadequate levels of support, this heavy reliance often resulted in feelings of exhaustion and exacerbation of pre-existing, or new, health conditions. One mother, for example, described her experience as follows:

> They [family members] wanted to be around her [the child], they just didn't want to, um, touch her, because they thought she would break or, um, they-she would have a [respiratory] episode. And every-and if someone was holding her and they-and she, um, had a-an episode, then they would immediately hand her to me. Including my husband, including my- yeah, everyone. I was literally stuck to her 24/7. For two years. I gained a lot of weight.

This comment highlights how all-consuming caring for a preterm child can be during infancy when the child's health conditions are still being managed. In addition, the overall adverse impact on the mothers' general health was identified. It is important to note, however, that as children grew into adolescence, most mothers (90%) reported resolving or learning to adapt to their child's physical health conditions. For mothers reporting little support, however, high levels of stress were still common. This was particularly emphasized by mothers whose child had ongoing, or unmanaged, neurodevelopmental or health conditions.

While all mothers emphasized the importance of their role as caregivers, what this role entailed varied by the child's abilities. Mothers of children with mainly neurotypical abilities emphasized teaching their child independence, the need for normalcy, and transitioning into adulthood. In contrast, mothers of children with moderate to severe neurodevelopmental conditions emphasized the need to adapt to their child's abilities, which often involved planning for ongoing care beyond childhood. A quote from a mother of a child with an intellectual disability, for example, summarizes how mothers matched their own expectations to their child's unique capabilities:

> My fear is how he going to be when he gets a little bit older. He wants to go to college. He said he- 'I'm going to college'; he'll say, 'I'm going to state.' He got high expectations for himself, and I do not discourage that. But I know he's going to have some, limitations- he cannot do that. So. he might have to do a tech. Now this is going on right now. Now as he gets older-but right now, I don't see that. He's just going to have to have baby steps in life.

In addition, advocacy skills, and its focus, varied among mothers. Overall, however, most mothers described advocacy as a method of obtaining services for their child, particularly within the school system. Mothers with a professional background in an academic setting, for example, seemed better able to navigate the school system and work alongside school personnel to create personalized education plans that provided their child with academic resources and aid. In contrast, mothers who identified a need for services, but were unfamiliar with the school system, expressed frustration at the numerous barriers they experienced attempting to obtain resources or classroom accommodations for their child.

Additionally, for mothers of children with milder neurodevelopmental symptoms, there was a balancing act of needs. While mothers wanted their child to have the help they needed, they had to

Table 2
Description of Caregiver/Child Demographic Characteristics.

Caregiver	Maternal Education	Maternal Annual Income	Child/Children's Race/Ethnicity	Child/Children's Diagnosis(es)
1. Mother	College	$30–59 k	White	None
2. Mother	College	$60–99 k	African American	None
3. Mother	High School	$30–59 k	African American	Asthma, Speech Delay, Learning Disability & Intellectual Disability
4. Mother	Some College	<$30 k	African American	Asthma
5. Mother	High School	<$30 k	White	Asthma, ADD/ADHD†, Autism/Pervasive Developmental Disorder, Developmental Delay, Speech Delay, Learning Disability & Intellectual Disability
6. Mother	College	$60–99 k	White	ADD/ADHD†, Autism/Pervasive Developmental Disorder, Tics, Learning Disability & Intellectual Disability
7. Mother	Graduate School	$100 k+	White	Developmental Delay, Speech Delay & Learning Disability
8. Mother	College	<$30 k	White	ADD/ADHD†, Developmental & Speech Delay
9. Mother	Less than High School	<$30 k	Black	ADD/ADHD†, Cerebral Palsy & Oppositional Defiant
10. Mother	Some College	$60-90 k	White	Asthma, CLD‡/Respiratory Disorder, ADD/ADHD†, Developmental Delay, Speech Delay & Learning Disability
11. Mother	Graduate School	$60-99 k	White	ADD/ADHD†, Developmental Delay, Speech Delay & Learning Disability
12. Mother	Some College	<$30 k	Black	Asthma
13. Mother	College	$60–99 k	Black	Speech Delay
14. Mother	College	$60–99 k	White	None
15. Mother	Graduate School	$100 k+	White	Asthma, CLD‡/Respiratory Disorder, Cerebral Palsy, Developmental Delay, Speech Delay, Learning Disability & Epilepsy/Seizures
16. Mother	High School	<$30 k	Black	Asthma, ADD/ADHD†, Developmental Delay & Speech Delay
17. Mother	Less than High School	<$30 k	Hispanic/Latinx	None
18. Mother	Some College	$30–59 k	White	CLD‡/Respiratory Disorder, Developmental Delay, Speech Delay & Intellectual Disability
19. Mother	College	$30–59.k	White	None
20. Mother	High School	Unknown	White	Twin 1: Asthma, CLD†/Respiratory Disorder & Developmental Delay Twin 2: ADD/ADHD† & Learning Disability

†ADD/ADHD: Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder ‡CLD: Chronic Lung Disorder.

balance this with the need for having their child avoid stigma and appear neurotypical. The quote below summarizes how mothers struggled to balance these opposing needs:

> I was kind of thankful that she didn't have that label in school. Because she was able to stay in the mainstream classes even though she had an IEP [Individual Education Plan] and the PEP [Physical Education Program], um, she still was able to stay with her class. Cause it-it's definitely a struggle… whenever you're trying to incorporate a child with issues into a school and trying to make sure their life at the school is as normal as possible. Um, and the-but you want them to get all the help they can get while they're at the school so they can be as normal as they can.

Meeting these opposing needs occasionally resulted in failing to find and utilize resources, such as academic or behavioral support—a deficit of aid which some mothers believed a necessary sacrifice.

Individual coping factors identified by mothers included their spiritual beliefs, which were utilized to explain both incidences of positive and adverse health. When asked about factors that contributed to her child's positive health, one mother stated:

> It was like a struggle because I had to watch her [inhales deeply] leave and come back, leave and come back, you know, like she going to die and came-come back. I'm watching her with these blood transfusions. So, it was a bit stressful on me, but [inhales deeply] by me believing in God I knew she was going to be alright, with the pull through, with my faith. I had to keep my faith in her.

Additionally, mothers typically described their relationship with their child as a central component to their, and their child's, well-being. Mothers who felt they had a healthy, strong relationship with their child seemed to emphasize their child's personal strengths. For example, despite acknowledging her daughter's social challenges, this mother also wanted to emphasize her strengths as described below:

> You know, from a, from a sort of mental/emotional thing, I think, you know, one of the best parts about her is..she sort of has the ability to make people feel good.

In contrast, mothers who expressed having an overly dependent, or distant, relationship with their child tended to focus on their child's weaknesses and/or their worry for the child's future.

> Not nobody else he relies on, only on me. You know. And I try, I try to make him stronger and… more mature and independent… So that's my problem, that's my worry.

This difference emphasizes the importance of the mother-child relationship and how vital it is for the mother-child dyad to find a balance between closeness and independence.

The child's contribution to well-being

It is important to note that despite any physical diagnoses their child was still living with, most mothers (95%) described their child as physically healthy and capable of engaging in physical activities. While commonly initiated by family members, the child's participation in sports or outdoor activities was seen by most mothers to contribute greatly to their child's overall well-being. Aside from sports, having hobbies such as anime (a style of Japanese film and television animation aimed at both children and adults), cooking, or electronic video games allowed the child to develop their own interests and gave the child a sense of independence and self apart from the caregiver. Even for children with a neurodevelopmental disorder, having a hobby allowed the child to begin taking steps toward independence.

In contrast, mothers varied in their description of their child's well-being. While some adolescents were described as isolated or showing symptoms of anxiety, others were described as happy and well-adjusted adolescents. Having age-appropriate interpersonal skills was seen as vital by most mothers to the promotion of psychological health and overall well-being in their child. Youth who were described as able

to appropriately engage with peers were viewed as having more success in having their emotional and social needs met. In contrast, when mothers described the child as having reduced interpersonal skills, they also expressed concern about the child's limited peer network. Some mothers noted the child's heavy reliance on themselves, or siblings, for social engagement and support. One mother summarized her concern and frustration about her child's limited social interactions, which she believed contributed to a sedentary lifestyle in the following quote:

> But if he goes with friends, they, you know, we got kids around here, some of them might be his age some not, he'll do it but other than that [NAME OF CHILD] stay right here under this house, up under me. He won't' care nothing about going outside playing. He'll' go out for a little while and he'll' come right back in this house.

These social limitations seem to not only affect the child's social well-being but have adverse implications for the child's physical and mental health.

The family unit

Mothers described the family unit as a key influence in their child's well-being, with family functioning and child well-being being interdependent. Having a supportive partner, even if they were divorced, seemed to greatly promote overall health and well-being in both the child and mother. Co-parenting was experienced by 45% of participants as a state where caregiving and management of household tasks was seen as a shared responsibility. In contrast, 55% of mothers described having limited, or no, support.

In addition to co-parenting, mothers described how spending time together and engaging in shared hobbies greatly contributed to the well-being of their child. When family members had a shared interest in sports, emphasis was often placed on the youth's ongoing involvement in physical activities. In contrast, for families who did not have shared interests, the child was often responsible for initiating their own physical activities and hobbies. Unfortunately, relying solely on the child's self-initiation often seemed to result in a largely sedentary lifestyle.

In addition to sports, shared family values, such as being socially engaged with their neighborhood or religious community, seemed to result in children who were more socially engaged. In contrast, families who were isolated or had little contact with members of their community seemed to have a reduced social network, often resulting in the mother turning to more formal resources for support.

Finally, the family's economic resources greatly affected the child's, and mother's, overall well-being. Economic resources were described as material resources or support from their job and/or extended family. Mothers who described themselves as economically stable expressed how relieved they were at being able to take time off without fear of losing their job. This was emphasized particularly during their child's early development, when mothers had to often address their child's ongoing medical needs. Additionally, extended family members often provided the family unit with financial, or material, support, as exemplified by this quote:

> Because, you know, not only are you dealing with those emotional, emotional stresses. (Inhales breath) but you're dealing with the financial side of it, too. You know, um, I didn't work, my husband was the only one working. So that was a challenge, um, just figuring out everything. You know, we sold everything down to the fact that we didn't sell our house, that was the only thing we had left. Um, we kept our house. And my mom loaned me a vehicle. So, my vehicle we could concentrate only on the kids [twins].

This quotation highlights how extended family members often addressed gaps in resources and/or aid. Therefore, when familial support is absent in families with limited economic resources, there was often adverse economic implications.

Theme II. Contributors & barriers at the community level

The second theme, which focused on contributors and barriers at the community level consisted of two subthemes: a) *Formal Sources of Support* and b) *Informal Sources of Support*. The participant's community was divided into formal and informal sources of support and, on occasion, barriers. Formal sources of support that mothers described as contributing to their child's well-being included the healthcare and school systems and religious institutions. Informal sources of support included friends and the family's social and/or physical community.

Formal sources of support

Conflicting accounts were given in terms of the contribution the healthcare and school system had to the child's well-being. While both were seen as vital in their child's physical and psycho-social development, there were many barriers experienced by caregivers in accessing services provided by these institutions. In terms of the healthcare system, all children received care at the same tier one institution from birth to discharge, with most (95%) mothers conveying an overall positive experience in the care received.

Once discharged, however, the healthcare experience varied based on the healthcare provider's location and the mother's advocacy skills. While most seemed to be satisfied with their child's overall healthcare experience, some mothers voiced frustrations in accessing specialized care. This was particularly relevant for mothers whose child received care in rural areas, as expressed by one mother:

> Well, at first, there were issues with the healthcare team, because, um, I-we were going to my older daughter's doctor's office [in rural county]. And I had specifically asked them beforehand if they had any experience with preemies and stuff like that... and they said yes. And, um, whenever it came down to it, and she ha-had, she had pneumonia. She-she'd gotten pneumonia one time, um, not-it was like, uh, three months after she came home. Um, I called their emergency line and they just kept telling me to... keep switching off her, um, Motrin and Tylenol and stuff and I was telling her she wasn't, um, breathing right and stuff, and then I just took her to the hospital. I just took her to the emergency room. And if I hadn't, the-the doctors there told me that if I hadn't had done that, she could have been dead within 24 h. And so, the fact that the doctor's office that I had started her with didn't recognize that with her being a preemie and having respiratory issues and everything. I feel like they fell down on their duties.

Overall, mothers often felt responsible for identifying and navigating the healthcare system without little to any help.

Like the healthcare system, the school system was seen as a core institution in the child's life. Additionally, their experience also varied by location and the mother's ability for advocacy. Interestingly, a mother's ability to advocate within the school system was not solely dependent on her educational background. Instead, participants from varied educational backgrounds were often forced to learn advocacy skills. Within this experience, participants often described their frustration with the school failing to recognize their child's academic needs, sometimes regardless of a formal medical diagnosis. One participant, for example, highlighted how difficult it can be for caregivers to ensure their child is receiving the appropriate level of accommodations:

> I try to tell them even with an IEP, I try to tell him, they not doing like they should be with him in school. So, I have been going toe-to-toe with them teachers...so we had to do a-a conference call which I didn't like, because I don't have to tell you, but you want to ask questions and... that's when I start most of my concern and my anger toward them how they were doing him...

In addition to the school and the healthcare system, mothers emphasized the importance early intervention played in their child's

development. Specifically, services such as head start, speech and occupational therapy were emphasized. Based on their accounts, mothers believed that by participating in these activities, their child was able to begin reaching milestones much earlier than they had anticipated. In addition, while not considered interventions, daycare and pre-Kindergarten programs allowed their child to socialize and begin developing interpersonal skills, something which mothers emphasized was vital to their child.

Informal sources of support

In addition to formal sources of support, informal sources of support included having friends or extended family that lived in proximity. Mothers emphasized how their family and friends provided a safe environment for their child to practice socializing without fear of being ostracized. In addition, the family's social community often provided both material and nonmaterial resources to the child and family—such as childcare, someone to talk to, or even financial help. For families who had a very limited social support network, mothers often expressed feeling overwhelmed and lonely. Thus, an extended social support network of family and friends was described as vital to the well-being of both the child and the caregiver.

In addition to the social environment, mothers described how the neighborhood in which the child lived played a crucial role in child and family well-being. While most participants described their physical neighborhood as safe, mothers who lived in areas of high crime activity described being fearful and engaging in protective measures such as limiting—physical activities outside of the home or the formation of peer relationship with neighborhood children. While effective in keeping the child physically safe, these protective strategies often resulted in the child becoming more isolated and sedentary.

Religious institutions provided both formal and informal sources of support for the caregiver, child, and family. While formal resources included religious and spiritual guidance and support, informal sources of support included providing the caregiver and child with a place to form friendships and create a social support network.

Discussion

For this paper we explored the caregivers' perspective about aspects of their social, community, and family environment which they believed affected their extremely preterm child's development and well-being. While prior studies have explored caregivers' experiences during early childhood for extremely preterm child (Granero-Molina et al., 2019; Schuetz Haemmerli et al., 2020), very limited information currently exists that looks beyond this developmental period to adolescence. Additionally, while we were unable to compare by variations in health, we were able to identify in-depth environmental characteristics that affect extremely preterm children's overall well-being. Something which has received little study in preterm literature, with most studies focusing on general factors like maternal education or income (Bangma et al., 2018; Bangma et al., 2019; Benavente-Fernández et al., 2019).

Our findings indicated that the role of the mother is essential to EP children's development and well-being. However, it is important to note that we were only able to interview mothers, therefore, a full understanding of the role significant others play was not possible with the current data. Overall, mothers emphasized their role as the primary caregiver, and advocate, of their EP child. However, this heavy reliance on self often resulted in maternal exhaustion and exacerbation of pre-existing, or new, health conditions. These results align with the finding that mothers of EP children experience increased stress during their EP child's early childhood (O'Donovan & Nixon, 2019; Schuetz Haemmerli et al., 2020). The adverse effects to physical and mental health, however, have not been as well-studied and was an important finding in this study—indicating a vital need for clinicians and healthcare professionals to provide ongoing support to this vulnerable population.

In addition, while mothers emphasized their role as caregivers and advocates, the type of care or advocacy provided varied based on the child's development and health. For children with no, or milder, developmental conditions, the need for normalcy was emphasized by mothers. For this group, most behavioral interventions seemed to occur early in childhood and decreased as the child's health needs decreased. This occasionally resulted in EP children with learning or developmental differences having limited academic or behavioral resources to rely on later in childhood—a deficit which for some resulted in difficulties in school or job maintenance once they graduated high school. This aligns with literature indicating that compared to term-born adults, adults born preterm have been found to be less likely to be employed or have higher educational qualifications, even after controlling for maternal socioeconomic status (Bilgin et al., 2018; Crump et al., 2019).

For EP youth, participation in sports or outdoor activities was expressed by participants as essential to the development of their child's physical health. It is important to note, however, that while occasionally self-motivated, many of these EP youth's involvement in physical activities was initially motivated by a family member, with many caregivers emphasizing the importance of a specific sport or outdoor activity. This aligns with literature in the general population indicating that participation in organized physical activity early in life is associated with higher rates of physical activity in adolescence (Spiegler et al., 2019). Interestingly, most of the EP youth seemed to be self-motivated to pursue their own hobbies, which allowed them a sense of self. These findings parallel literature among individuals with disabilities that emphasize how vital having a hobby can be in developing a sense of self and increasing self-esteem (Patterson, 2001; Specht et al., 2002).

At the community level, the healthcare and school system were seen as both a facilitator and barrier to well-being for EP individuals, with many participants finding it difficult to obtain services without extensive knowledge of both systems. These results coincide with literature indicating how complex it can be for caregivers to access services within the school and healthcare system (Boshoff et al., 2016; Magaña et al., 2013). Finally, a family's social network and physical environment were seen as vital to individual and family well-being. While a supportive and extensive social network provided families with both emotional and material sources of support, participants with limited social networks reported feeling isolated and often overwhelmed by their child's needs. This isolation was particularly felt during their child's early childhood, when the child's medical needs tended to be the highest.

Like social networks, the physical environment in which the family lived greatly affected individual and family well-being. While most families lived in safe neighborhoods, for those who did not, the threat often increased stress and made them take vigilant measures to ensure their child's safety. These behaviors often ran counter to what they believed would be best for their child's health—including limiting outdoor activity or limiting their child's peer network.

Strengths & limitations

This qualitative descriptive study addressed the limited knowledge that currently exists about conditions and dynamics at the family and community level that affect EP youth's development and well-being. Limitations of this study include the inability to fully explore how gender, race and/or culture affect these experiences. Literature has identified how race and/or ethnicity impacts the care and services EP children receives both in the NICU and in follow-up care (Beck et al. 2019; Sigurdson et al., 2018). For this study, we did not include enough participants from a black or Hispanic/Latinx background to fully explore this phenomenon. In addition, all participants were mothers, a limitation which fails to fully describe the experience of fatherhood. Therefore, more research is needed in fully exploring how well-being at the family and community level is affected by gender, race and/or ethnicity.

Research & practice implications

We were able to identify conditions and dynamics at the family and community level that have the potential to affect an extremely preterm child's development and well-being. However, more research is needed to explore how these experiences vary based on racial/ethnic background and financial resources. Specifically, access to helpful resources such as early childcare, and safe outdoor areas, are closely associated with a family's financial resources.

Healthcare providers caring for these families must realize that not only are EP youth affected by prematurity, but caregivers are also deeply impacted both emotionally and physically by caring for the preterm infant as they mature. Therefore, it is essential that maternal and family care is also emphasized in community practice.

Conclusion

Overall, mothers of extremely preterm children are vital in the extremely preterm child's well-being. While there are individual level characteristics that contribute to well-being, a support structure at the family and community level is essential to the child and mother's well-being. Specifically, families with limited material and/or immaterial resources rely on formal institutions, such as the school and healthcare system for resources. Therefore, it is vital that personnel in these institutions ensure that their services are easily accessible for this vulnerable population group.

Credit authorship contribution statement

Crisma Emmanuel: Conceptualization, Methodology, Investigation, Writing – original draft. **Kathy A. Knafl:** Conceptualization, Writing – review & editing. **Sharron L. Docherty:** Conceptualization, Writing – review & editing. **Eric A. Hodges:** Writing – review & editing. **Janice K. Wereszczak:** Conceptualization, Resources, Writing – review & editing. **Julie V. Rollins:** Writing – review & editing. **Rebecca C. Fry:** Writing – review & editing. **T. Michael O'Shea:** Conceptualization, Writing – review & editing. **Hudson P. Santos:** Supervision, Conceptualization, Writing – review & editing.

Declaration of Competing Interest

No declaration of interest to report.

Acknowledgments

The authors would like to acknowledge the contributions of the ELGAN subjects and the ELGAN subjects' families. Additionally, they would like to acknowledge funding received from the National Institute of Nursing Research (R01NR019245), the National Institutes of Health Office of the Director (UG3OD023348), and the Rita & Alex Hillman Foundation. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health or the Rita & Alex Hillman Foundation.

References

Armat, M. R., Assarroudi, A., Rad, M., Sharifi, H., & Heydari, A. (2018). Inductive and deductive: Ambiguous labels in qualitative content analysis. *The Qualitative Report*, *23*(1), 219–221.

Bangma, J. T., Kwiatkowski, E., Psioda, M., Santos, H. P., Jr., Hooper, S. R., Douglass, L., ... Fry, R. C. (2018). Assessing positive child health among individuals born Extremely preterm. *The Journal of Pediatrics*, *202*, 44–49.e44. https://doi.org/10.1016/j.jpeds.2018.06.037.

Bangma, J. T., Kwiatkowski, E., Psioda, M., Santos, H. P., Jr., Hooper, S. R., Douglass, L., ... Investigators, E. (2019). Early life antecedents of positive child health among 10-year-old children born extremely preterm. *Pediatric Research*, *86*(6), 758–765. https://doi.org/10.1038/s41390-019-0404-x.

Barnett, M. L., Tusor, N., Ball, G., Chew, A., Falconer, S., Aljabar, P., ... Counsell, S. J. (2018). Exploring the multiple-hit hypothesis of preterm white matter damage using diffusion MRI. *NeuroImage. Clinical*, *17*, 596–606. https://doi.org/10.1016/j.nicl.2017.11.017.

Benavente-Fernández, I., Synnes, A., Grunau, R. E., Chau, V., Ramraj, C., Glass, T., ... Miller, S. P. (2019). Association of socioeconomic status and brain injury with neurodevelopmental outcomes of very preterm children. *JAMA Network Open*, *2*(5), e192914. https://doi.org/10.1001/jamanetworkopen.2019.2914.

Bilgin, A., Mendonca, M., & Wolke, D. (2018). Preterm birth/Low birth weight and markers reflective of wealth in adulthood: A Meta-analysis. *Pediatrics*, *142*(1), Article e20173625. https://doi.org/10.1542/peds.2017-3625.

Boshoff, K., Gibbs, D., Phillips, R. L., Wiles, L., & Porter, L. (2016). Parents' voices: "why and how we advocate". A meta-synthesis of parents' experiences of advocating for their child with autism spectrum disorder. *Child: Care, Health and Development*, *42*(6), 784–797. https://doi.org/10.1111/cch.12383.

Crump, C., Winkleby, M. A., Sundquist, J., & Sundquist, K. (2019). Prevalence of survival without major comorbidities among adults born prematurely. *Jama*, *322*(16), 1580–1588. https://doi.org/10.1001/jama.2019.15040.

Doyle, L., McCabe, C., Keogh, B., Brady, A., & McCann, M. (2020). An overview of the qualitative descriptive design within nursing research. *Journal of research in nursing : JRN*, *25*(5), 443–455. https://doi.org/10.1177/1744987119880234.

Doyle, L. W., Cheong, J. L., Burnett, A., Roberts, G., Lee, K. J., & Anderson, P. J. (2015). Biological and social influences on outcomes of extreme-preterm/Low-birth weight adolescents. *Pediatrics*, *136*(6), e1513–e1520. https://doi.org/10.1542/peds.2015-2006.

Elo, S., & Kyngäs, H. (2008). The qualitative content analysis process. *Journal of Advanced Nursing*, *62*(1), 107–115. https://doi.org/10.1111/j.1365-2648.2007.04569.x.

Evans, T., Whittingham, K., Sanders, M., Colditz, P., & Boyd, R. N. (2014). Are parenting interventions effective in improving the relationship between mothers and their preterm infants? *Infant Behavior and Development*, *37*(2), 131–154. https://doi.org/10.1016/j.infbeh.2013.12.009.

Family Caregiver Alliance (2021). Definitions. Retrieved September 24, 2021 fromhttps://www.caregiver.org/resource/definitions-0/.

Gonçalves, J. L., Fuertes, M., Alves, M. J., Antunes, S., Almeida, A. R., Casimiro, R., & Santos, M. (2020). Maternal pre and perinatal experiences with their full-term, preterm and very preterm newborns. *BMC Pregnancy and Childbirth*, *20*(1), 276. https://doi.org/10.1186/s12884-020-02934-8.

Granero-Molina, J., Fernández Medina, I. M., Fernández-Sola, C., Hernández-Padilla, J. M., Jiménez Lasserrotte, M., & López Rodríguez, M. D. M. (2019). Experiences of mothers of Extremely preterm infants after hospital discharge. *Journal of Pediatric Nursing*, *45*, e2–e8. https://doi.org/10.1016/j.pedn.2018.12.003.

Harding, C., Levin, A., Crossley, S. L., Murphy, R., & van den Engel-Hoek, L. (2019). Effects of early communication intervention on speech and communication skills of preterm infants in the neonatal intensive care unit (NICU): A systematic review. *Journal of neonatal nursing : JNN*, *25*(4), 177–188. https://doi.org/10.1016/j.jnn.2019.04.004.

Hennink, M. M., Kaiser, B. N., & Marconi, V. C. (2016). Code saturation versus meaning saturation: How many interviews are enough? *Qualitative Health Research*, *27*(4), 591–608. https://doi.org/10.1177/1049732316665344.

Holditch-Davis, D., Santos, H., Levy, J., White-Traut, R., O'Shea, T. M., Geraldo, V., & David, R. (2015). Patterns of psychological distress in mothers of preterm infants. *Infant Behavior and Development*, *41*, 154–163. https://doi.org/10.1016/j.infbeh.2015.10.004.

Janevic, T., Zeitlin, J., Auger, N., Egorova, N. N., Hebert, P., Balbierz, A., & Howell, E. A. (2018). Association of Race/ethnicity with very preterm neonatal morbidities. *JAMA Pediatrics*, *172*(11), 1061–1069. https://doi.org/10.1001/jamapediatrics.2018.2029.

Joseph, R. M., O'Shea, T. M., Allred, E. N., Heeren, T., Hirtz, D., Jara, H., ... Kuban, K. C. (2016). Neurocognitive and academic outcomes at Age 10 years of Extremely preterm newborns. *Pediatrics*, *137*(4). https://doi.org/10.1542/peds.2015-4343.

Joseph, R. M., O'Shea, T. M., Allred, E. N., Heeren, T., & Kuban, K. K. (2018). Maternal educational status at birth, maternal educational advancement, and neurocognitive outcomes at age 10 years among children born extremely preterm. *Pediatric Research*, *83*(4), 767–777. https://doi.org/10.1038/pr.2017.267.

Kuban, K. C., Joseph, R. M., O'Shea, T. M., Allred, E. N., Heeren, T., Douglass, L., ... Extremely Low Gestational Age Newborn Study, I (2016, Jun). Girls and boys born before 28 weeks gestation: Risks of cognitive, behavioral, and neurologic outcomes at Age 10 years. *The Journal of Pediatrics*, *173*(69–75), Article e61. https://doi.org/10.1016/j.jpeds.2016.02.048.

Kuzniewicz, M. W., Wi, S., Qian, Y., Walsh, E. M., Armstrong, M. A., & Croen, L. A. (2014). Prevalence and neonatal factors associated with autism spectrum disorders in preterm infants. *The Journal of Pediatrics*, *164*(1), 20–25. https://doi.org/10.1016/j.jpeds.2013.09.021.

Long, K. M., McDermott, F., & Meadows, G. N. (2018). Being pragmatic about healthcare complexity: Our experiences applying complexity theory and pragmatism to health services research. *BMC Medicine*, *16*(1), 94. https://doi.org/10.1186/s12916-018-1087-6.

Luu, T. M., Rehman Mian, M. O., & Nuyt, A. M. (2017). Long-term impact of preterm birth: Neurodevelopmental and physical health outcomes. *Clinics in Perinatology*, *44*(2), 305–314. https://doi.org/10.1016/j.clp.2017.01.003.

Magaña, S., Lopez, K., Aguinaga, A., & Morton, H. (2013). Access to diagnosis and treatment services among latino children with autism spectrum disorders. *Intellectual and Developmental Disabilities*, *51*(3), 141–153.

Mathewson, K. J., Chow, C. H., Dobson, K. G., Pope, E. I., Schmidt, L. A., & Van Lieshout, R. J. (2017). Mental health of extremely low birth weight survivors: A systematic review and meta-analysis. *Psychological Bulletin*, *143*(4), 347–383. https://doi.org/10.1037/bul0000091.

Morgan, A. S., Mendonça, M., Thiele, N., & David, A. L. (2022). Management and outcomes of extreme preterm birth. *BMJ*, *376*, e055924. https://doi.org/10.1136/bmj-2021-055924.

Morse, J. M. (2015). Critical analysis of strategies for determining rigor in qualitative inquiry. *Qualitative Health Research, 25*(9), 1212–1222. https://doi.org/10.1177/1049732315588501.

Office, E., Kigenyi, T., Sondik, E., Pronk, N., Kleinman, D. V., Blakey, C., ... Brewer, K. H. (2021). Healthy People 2030 Leading health indicators and overall health and well-being measures: Opportunities to assess and improve the health and well-being of the nation. *Journal of Public Health Management and Practice, 27*(Suppl. 6), S235–S241. https://doi.org/10.1097/PHH.0000000000001424.

O'Donovan, A., & Nixon, E. (2019). "weathering the storm:" Mothers' and fathers' experiences of parenting a preterm infant. *Infant Mental Health Journal, 40*(4), 573–587.

O'Shea, T. M., Allred, E. N., Dammann, O., Hirtz, D., Kuban, K. C. K., Paneth, N., ... Investigators, E. s (2009). The ELGAN study of the brain and related disorders in extremely low gestational age newborns. *Early Human Development, 85*(11), 719–725. https://doi.org/10.1016/j.earlhumdev.2009.08.060.

Palinkas, L. A., Horwitz, S. M., Green, C. A., Wisdom, J. P., Duan, N., & Hoagwood, K. (2015). Purposeful sampling for qualitative data collection and analysis in mixed method implementation research. *Administration and Policy in Mental Health, 42*(5), 533–544. https://doi.org/10.1007/s10488-013-0528-y.

Patterson, I. (2001). Serious leisure as a positive contributor to social inclusion for people with intellectual disabilities. *World Leisure Journal, 43*(3), 16–24. https://doi.org/10.1080/04419057.2001.9674234.

Patton, M. Q. (1990). Designing qualitative studies. *Qualitative evaluation and research methods* (pp. 169–186). Sage.

Penagoch, J., Vidal, R., Gómez-Lumbreras, A., Hermosilla, E., Riera, L., Cortés, J., ... Morros, R. (2021). Prematurity and ADHD in childhood: An observational register-based Study in Catalonia. *Journal of Attention Disorders, 25*(7), 933–941. https://doi.org/10.1177/1087054719864631.

Richardson, G. E. (2002). The metatheory of resilience and resiliency. *Journal of Clinical Psychology, 58*(3), 307–321. https://doi.org/10.1002/jclp.10020.

Roberts, G., Burnett, A. C., Lee, K. J., Cheong, J., Wood, S. J., Anderson, P. J., & Doyle, L. W. (2013). Quality of life at Age 18 years after Extremely preterm birth in the post-surfactant era. *The Journal of Pediatrics, 163*(4), 1008–1013.e1001. https://doi.org/10.1016/j.jpeds.2013.05.048.

Rutten, B. P. F., Hammels, C., Geschwind, N., Menne-Lothmann, C., Pishva, E., Schruers, K., ... Wichers, M. (2013). Resilience in mental health: Linking psychological and neurobiological perspectives. *Acta Psychiatrica Scandinavica, 128*(1), 3–20. https://doi.org/10.1111/acps.12095.

Saigal, S. (2016). In their own words: Life at adulthood after very premature birth. *Seminars in Perinatology, 40*(8), 578–583. https://doi.org/10.1053/j.semperi.2016.09.010.

Sandelowski, M. (2000). Combining qualitative and quantitative sampling, data collection, and analysis techniques in mixed-method studies. *Research in Nursing & Health, 23*(3), 246–255.

Sandelowski, M. (2010). What's in a name? Qualitative description revisited. *Research in Nursing & Health, 33*(1), 77–84. https://doi.org/10.1002/nur.20362.

Schuetz Haemmerli, N., Lemola, S., Holditch-Davis, D., & Cignacco, E. (2020). Comparative evaluation of parental stress experiences up to 2 to 3 years after preterm and term birth. *Advances in Neonatal Care, 20*(4), 301–313. https://doi.org/10.1097/anc.0000000000000714.

Serenius, F. (2016). Neurodevelopmental outcomes among Extremely preterm infants 6.5 years after active perinatal Care in Sweden. *JAMA Pediatrics, 170*(10), 954–963. https://doi.org/10.1001/jamapediatrics.2016.1210.

Specht, J., King, G., Brown, E., & Foris, C. (2002). The importance of leisure in the lives of persons with congenital physical disabilities. *The American Journal of Occupational Therapy, 56*(4), 436–445. https://doi.org/10.5014/ajot.56.4.436.

Spiegler, J., Mendonca, M., & Wolke, D. (2019). Prospective Study of physical activity of preterm born children from Age 5 to 14 years. *The Journal of Pediatrics, 208*, 66–73.e67. https://doi.org/10.1016/j.jpeds.2018.12.013.

Taylor, G. L., & O'Shea, T. M. (2022). Extreme prematurity: Risk and resiliency. *Curr Probl Pediatr Adolesc Health Care, 101132*. https://doi.org/10.1016/j.cppeds.2022.101132.

Vik, M. H., & Carlquist, E. (2018). Measuring subjective well-being for policy purposes: The example of well-being indicators in the WHO "health 2020" framework. *Scandinavian Journal of Public Health, 46*(2), 279–286. https://doi.org/10.1177/1403494817724952.

Walsh, F. (2003). Family resilience: A framework for clinical practice. *Family Process, 42*(1), 1–18. https://doi.org/10.1111/j.1545-5300.2003.00001.x.