Voices of experience in periviable decision-making and artificial placenta technology

Boer, A.H.A. de

Citation

Boer, A. H. A. de. (2025, July 1). *Voices of experience in periviable decision-making and artificial placenta technology*. Retrieved from https://hdl.handle.net/1887/4252056

Chapter 3

Perspectives of extremely prematurely born adults on what to consider in prenatal decision-making: a qualitative focus group study

Angret de Boer, Lien De Proost, Marieke de Vries,
Marije Hogeveen, E.J.T. (Joanne) Verweij, Rosa Geurtzen

Archives of Disease in Childhood: Fetal and Neonatal Edition, 2024

Abstract

Objective: A shared decision-making (SDM) approach is recommended for prenatal decisions at the limit of viability, with a guiding role for parental values. People born extremely premature experience the consequences of the decision made, but information about their perspectives on prenatal decisions is lacking. Therefore, this study aims to describe their perspectives on what is important in decision-making at the limit of viability.

Design: Semi-structured focus group discussions were conducted, recorded, and transcribed verbatim. The data were independently analysed by two researchers in Atlas.ti.

Results: Four focus groups were conducted in the Netherlands, with 5-6 participants each, born between $24^{0/7}$-$30^{0/7}$ weeks gestation in the period between 1965-2002. Considering their personal life experiences and how their extremely premature birth affected their families, the participants reflected on decision-making at the limit of viability. Various considerations were discussed and summarized into the following themes: anticipated parental regret, the wish to look at the baby directly after birth, to give the infant a chance at survival, quality of life, long term outcomes for the infant and the family, and religious or spiritual considerations.

Conclusions: Insights into the perspectives of adults born extremely premature deepened our understanding of values considered in decision-making at the limit of viability. Results point out the need for a more individualized prediction of the prognosis and more extensive information on the lifelong impact of an extremely premature birth on both the infant and the family. This could help future parents and healthcare professionals in value-laden decision-making.

Background

When an extremely premature birth is imminent, a prenatal decision must be made whether to start early intensive care (EIC) or palliative comfort care (PCC).[1] Accurately predicting individual outcomes is difficult and the evaluation of the estimated prognosis is value laden.[2,3] Therefore, shared decision-making (SDM) is recommended which includes the elicitation and incorporation of parental values.[4] Various parental perspectives are known to be important in decision-making, such as long-term outcomes, survival and the impact on the family-system.[5] Parents may choose EIC to "*give a chance*" and to "*do everything possible*",[6,7] while others may choose PCC to "*avoid a life of challenges and potential handicaps*".[8,9]

Adults born extremely premature experience the consequences of their extremely premature birth[10] and witness the impact on their families.[11] Although the importance of reporting long-term medical, psychological and social outcomes are being increasingly recognized,[12] the perspectives of these individuals on the prenatal decision-making process are still unknown.[5] Although parents are the surrogate decision-makers for their infant, understanding the perspectives of adults who were born prematurely is valuable in that it represents how the actual patients ultimately reflect on this decision. To give voice to the individuals most affected by these decisions, this study aims to describe the perspectives of extremely premature born adults on what they believe is important in decision-making at the limit of viability. This may support healthcare professionals and future parents in SDM.

Methods

TINY-1-study

This qualitative study was conducted as part of the TINY project (Towards Individualized care for the Youngest) by researchers from the Leiden University Medical Centre; Erasmus Medical Centre, Rotterdam; and Radboud University Medical Center Nijmegen, in the Netherlands. Ethical approval was obtained from the Medical Ethics Committee Leiden-Den Haag-Delft (N21.147, November 2021). Topics of interests are (1) guidelines, (2) personalization, and (3) values in decision-making at the limits of viability. This article presents the results of TINY-1 on values important in decision-making.

Study design and participants

Focus group discussions were conducted, aiming for a limited number of participants in each group to create a safe environment but still facilitate discussion. Participants were recruited through the Dutch patient association Care4Neo and social media (e.g., LinkedIn). Participants included had to be born extremely premature, >18yr of age and able to verbally express themselves well. Individuals who were interested to participate contacted the researchers via email. Information and informed consent forms were sent. All individuals who signed up and met the inclusion criteria, were included in the study. After signing up, a questionnaire for background information followed to collect demographic and health-related information. Participants were divided into focus groups, ensuring diversity in age, gender and health status. An individual meeting was planned with each participant around one week before the focus group discussion to elaborate on the research project's background and aim, and to answer any questions. Information was shared, including the number of participants, the structure of the focus group discussions, the duration and other relevant logistical details. After the focus group discussions, an individual debriefing meeting was offered as aftercare to all because of the potential impact the discussions could have. We discussed how they participants experienced their participation in the focus group discussion. A 30-min meeting was held with 15 of the 23 participants, who all had a positive experience.

Data collection

Due to COVID-19 restrictions, the focus group discussions were held through video conferences. All were moderated by the same researchers LP and AB, with backgrounds in philosophy and bioethics (LP) and medicine (AB). At each session, a third researcher (neonatologist (RG) or obstetrician (JV)) was present to answer questions but not participate in the discussion.

An interviewguide was developed based on a review about what is known in literature on this subject.(5) It comprised (I) general instructions (aim, rules, and expectations) and (II) open-ended questions with follow-up probes for facilitating discussion (*appendix 1*). Treatment options at the limit of viability were explained, just as the aim to use an SDM-approach and incorporate parental values in decision-making. Open-ended questions were asked on important values, perspectives, or arguments to take into account when making a treatment decision. Finally, a figure representing an overview of parental perspectives gleaned from literature was shown and participants' comments were explored (*chapter 2*).(5)

Data analysis

The sessions were recorded and transcribed verbatim. Results were analysed and coded independently by two authors (AB, LP) in Atlas.ti, using thematic content analysis. The codebook and analysis were discussed and adjusted in multiple rounds until agreement was reached (AB, LP, RG, EV) (*appendix 2*). Findings were reported following the COREQ-criteria (*appendix 3*).(13) Every quote in the results section is accompanied by a number (session) and letter (participant).

Results

Focus group and participants' background

In January and February 2022, four focus group discussions were conducted with 5-6 participants per group (n = 23). With the aim of diversity each group was composed of participants with differences in the year of birth, gender and the self-reported long-term consequences. Characteristics and self-identified long-term consequences were reported in *Table 1* and *2A*. The participants in this study were born between $24^{0/7}$-$30^{0/7}$-weeks of gestation in the period between 1965 and 2002. The differences in outcome of our participants, born within a 37-year time span, could be partially explained by the range of potential outcomes of extreme prematurity but also by differences in neonatal treatment over time. Since the major advances in management of extremely premature infants, the long-term consequences of our participants are also reported broken down by their year of birth in *Table 2B*. No significant differences in experienced consequences are observed between the groups. Most of them experienced challenges of being born extremely premature influencing their lives as adults. Various consequences were reported, such as brain injury, cerebral paresis, bronchopulmonary dysplasia (BPD), visual impairment and hearing loss, as shown in *Table 2A* and *2B*. In some of these cases, the experienced consequences led to (e.g.) inability to work. Despite these consequences, most participants expressed how '*they got lucky*', and that '*it could have been worse*'. They were aware that other individuals born at the same gestational age suffered from more severe complications or did not survive.

Table 1 Self-reported sociodemographic and clinical characteristics of the participants.

Patient characteristic	N = 23
Participants	
Man	5
Woman	18
Born at gestational age	
24^{+0}-25^{+6}	3
26^{+0}-27^{+6}	13
28^{+0}-30^{+0}	7
Born between	
1965-1980	4
1981-1990	9
1991-2002	10
Education	
Secondary school / Secondary vocational education	5
Higher professional education	10
University education	8
Religion	
Christian	6
No religion	17
One of multiples	
Yes (twins)	3
No	20
Location premature birth	
Academic hospital	14
General hospital	6
Foreign hospital / at home	3
Long-term consequences of premature birth	
Yes	15
Possibly	2
No	6

Table 2A Self-identified and reported long-term consequences of their extreme premature birth

Consequences	N = 17
Cognitive	
Cognitive / learning disability / problems at school / brain injury / memory disorders	5
(Neuro)motor function	
Problems with moving (due to hypo/hypertonia) / cerebral paresis	9
Psychosocial	
Concentration problems / ADHD*	8
(symptoms of) Autism Spectrum Disorder	2
Other psychosocial problems	1
Physical	
Vision / hearing problems	7
Respiratory problems	7
Reduced immunity / susceptible to infection	5
Easily fatigued	5
Growth restriction	4
Eating/feedings problems	4
Intestinal problems	3
Other self-described consequences:	
Feeling misunderstood, attachment problems, anxiety disorder / performance anxiety, insecure / the need to proof themselves, stimulus processing problems	

*ADHD = Attention Deficit Hyperactivity Disorder

Important considerations in decision-making at the limit of viability

The participants in this study agreed with the SDM-approach as recommended in clinical guidelines and emphasized the importance of involving parents and their values in the decision-making process. Participants' explorations on values important in decision-making were multidimensional, reflecting on factual risk information as well as feelings and assumptions about care for extremely premature infants. The following themes were identified: anticipated regret or guilt, looking at the child after birth, giving a chance at survival, quality of life, consequences of EIC-treatment and religion and spirituality.

Table 2B Self-identified and reported long-term consequences of participants’ extreme premature birth by year of birth

Year of birth	Gestational ages	Reported long-term consequences of participants born in the reported period of time
<1980[1]	$26^{+?}$, $27^{+?}$, 30^{+0}	Cognitive / learning disability / problems at school / brain injury / memory disorders Problems with moving Reduced immunity / susceptible to infection Respiratory problems Intestinal problems (symptoms of) Autism Spectrum Disorder Stimulus processing problems Attachment problems
1980-1990[2]	$26^{+?}$ (2), 26^{+3}, 26^{+4}, 27^{+0}, $27^{+?}$, 28^{+0}, $28^{+?}$, 28^{+5}, 29^{+0}	Cognitive / learning disability / problems at school / brain injury / memory disorders Problems with moving/ Cerebral paresis Easily fatigued Vision / hearing problems Eating/feedings problems Respiratory problems Intestinal problems Reduced immunity / susceptible to infection Growth restriction (symptoms of) Autism Spectrum Disorder Feeling misunderstood Insecure / the need to proof themselves *Two persons born between 1980 and 1990 reported no consequences.*
1991-2002[3]	24^{+6}, 25^{+0}, 25^{+3}, 26^{+0}, $27^{+?}$ (2), 27^{+0}, 27^{+3}, $28^{+?}$, 29^{+6}	Cognitive / learning disability / problems at school / brain injury / memory disorders Problems with moving Vision / hearing problems Growth restriction Respiratory problems Reduced immunity / susceptible to infection *Four persons born in 1991 or later reported no consequences.*

[1] 1970-80: Advances in the respiratory management of the premature infant; CPAP, discovery of application of prenatal corticosteroids(30)

[2] 1980-90: First successful administration of surfactant to a newborn, family centered care expanded, Nitric Oxide for persistent pulmonary hypertension of the newborn (PPHN)(30)

[3] Before 2010, the Dutch threshold of viability was at 25 weeks.

Anticipated regret or guilt

Participants imagined that the decision on care for extremely premature infants may be influenced by anticipated regret or guilt, particularly in choosing EIC-treatment over PCC, as one participant noted: palliative care cannot be undone”. They acknowledged the uncertainty surrounding individual outcomes and prognosis, and the difficulty parents may have in making this decision. Participants sympathized with their parents for being in the situation of imminent extremely premature birth, certainly not blaming them for the decision they made.

Box 1: Quotes 'Anticipated regret or guilt'

"I can also imagine that some parents may feel guilty if an infant does not come out of it alive or at least not unscathed." (4A)

"As a parent, you cannot make a wrong choice. You do what you think is right." (2D)

Looking at the child after birth

In each session, participants suggested to make the decision based on the infant's condition immediately after birth. They emphasized the importance to consider EIC if the infant showed a will to live. Participants opted to look for signs such as movement, crying or discomfort.

Box 2: Quotes 'Looking at the child after birth'

"Look at movements, but also how comfortable the baby is. (...) How does the baby breathe in life? (...)" (4C)

"The activity of the infant, so whether it is moving or floppy. (...) whether it is trying to stay alive. I think you can tell a lot from that. (...)" (4A)

Giving a chance at survival

Participants imagined parents would typically want their infant to survive and to initiate treatment when there was a chance or hope on survival. They expressed that parents may want to seize the chance to see their infant grow up. However, participants considered it important that parents are informed about what an infant must go through to survive after extremely premature birth and take this into account. They emphasized that survival should not be pursued at all costs, and if the infant is not viable or the treatment is disproportional, palliative care should be considered. So, participants discussed that parents should be counselled about potential later decision moments regarding withdrawal of EIC-treatment if things changed for the worse.

Box 3: Quotes 'Giving a chance at survival'

"...a physician should really say what the chances of survival are, but maybe it is also good for parents to know what it takes to survive. (...) [My parents] found it quite difficult to see what I had to go through. They were not told what it would take" (3A)

"If your baby suffers severely and it becomes very heavy to endure, [the healthcare professionals] supposedly will not say: we will continue to the bitter end." (2A)

Quality of life

Quality of life (Qol) was an important theme discussed by the participants, but difficult to define. They were unsure how to determine what a good Qol is and by whom this should be determined. However, according to one participant QoL should not be determined based on potential physical handicaps.

Box 4: Quotes 'Quality of life'

"I find 'quality of life' a bit difficult. I agree with the words. (...) [but], what is quality of life?" (3D)

"*Why, who decides when life is fun or valuable? That is indeed very personal.*" (1B)

"*What you often hear people say, one of my great annoyances, is: "As long as the child can walk." Then my thoughts are: are you going to measure happiness or a child's happiness in life by what someone can physically do*?" (1A)

Consequences of early intensive care treatment

Overall, the consequences of EIC were mostly discussed in terms of lifelong consequences for the infant and the impact on the family. Several personal anecdotes and stories were shared while this topic was discussed.

Long-term consequences for the extremely premature infant

Concerns were raised about the underestimation of the consequences of being born extremely premature and the effects of being admitted to the NICU. It was agreed upon that parents should be well informed about all the long-term consequences, including physical and psychosocial challenges both in childhood and beyond, and the great impact it could have on life. Participants felt as if they were living in survival mode since their birth and their capabilities were often overestimated. One participant was at an age where she compared herself with others building a future, knowing she could never do or achieve the same as them.

From their perspective, more research is needed on long-term consequences, particularly the psychosocial aspects and the consequences in adulthood, so parents can be informed in more depth about the prognosis of their child.

Box 5: Quotes 'Long-term consequences for the extremely premature infant'

"A premature birth has a significant impact, psychological and emotional (...), it would be good to know that beforehand" (1F)

"I think, certainly for parents, long-term effects are not really about when the child is 30, 40, 50 years old, but more about when the child is a child?" **(1A)**

*"I did suffer from a minor motor disability afterwards, which also limited my social development, because I was less able to participate in a team sport, such as football. I certainly experienced a limitation during my high school days, but also during my student days. I could not always participate and keep up, so I missed a lot of social activities."***(2B)**

"The survival mode is always on, meaning constant turmoil in my head. (...) Not controlling all the stimuli, causing that I cannot keep up with and fully participate in society" **(2D)**

Impact on family

Furthermore, discussion about the impact of having an extremely premature infant on the family arose. The participants shared how their premature birth affected their family, especially their siblings, with examples like the absence of the parents during those first months of NICU-treatment influencing family life, how afraid a sister of one participant was to touch her during the first years, or the guilt one participant felt towards her siblings because of all the attention she got instead of her siblings. They emphasized the importance of considering the family context in the decision-making process, and of assessing the parental capacity to handle the consequences of having an extremely premature infant. Due to the potential impact on the family, participants thought social support should be addressed during counselling. Besides, parents' financial situation should be taken into consideration, as they may have to financially support their child in the future.

Overall, participants emphasized that while family impact should be considered during the decision-making process, this should not be the ultimate deciding factor. Rather, it should be discussed during counselling to prepare parents for the potential impact of having an extremely premature infant and to assess their needs for support.

Box 6: Quotes 'Impact on family'

"As my mother says to this day: "You are and will remain a child I worry about". And [my two sisters] aren't. I feel badly about that for them (...) It feels like the attention mostly goes to me, while my sisters deserve it just as much." (2D)

"at such a moment you may forget that it is also really intense mentally for yourself. That you also should be able to handle it, as a parent." (3D)

"...you also should ask - in both the positive and negative scenarios - what kind of social support there is, because you just can't do it alone and you just need your resources." (1C)

"I am lucky my parents can financially support me, because I cannot pay for my living on my own due to the medical support I need." (2D)

Religion and spirituality

The role of spirituality was expressed as an important factor. The role for religion was only speculated on, as it was not of personal importance for them.

Finally, after showing the presented figure of parental considerations extracted from literature (*chapter 2*), the participants mostly confirmed or expanded upon the perspectives that had already been discussed spontaneously.

Discussion

Our study offers unique insights into the perspectives of adults who are extremely premature born on what they themselves consider as being important in decision-making at the limit of viability. This group of adults born extremely premature has firsthand experience of the consequences of premature birth and witnessed the impact on their families. Participants suffered different gradations of consequences of their extremely premature birth. These outcomes ranged from experiencing one or more issues in domains such as (neuro) motor function, cognition, and psychosocial well-being, to experiencing no consequences whatsoever. Based on this diversity of experiences, they identified important issues to be considered during prenatal decision-making. Participants were grateful for being involved in research and for being heard, since they feel their expertise is underrepresented. Therefore, it is important to value and acknowledge their perspective, which could help in further policy development.

The perspectives of the adults born extremely premature placed unique emphasis on perspectives known from previous research. Our results substantiate (I) the wish to look at the child directly after birth to give a more individual prognosis, and the results provide new insights into (II) the impact on the family, and (III) the need for parents to consider the fact that extreme prematurity is a lifetime diagnosis with its challenges. These results can help in value-based counselling as part of personalized counselling and SDM.

First, the wish to "look at the child" and to use this information to decide, may reflect the hope for an uncertain and general prognosis to become a more individualized prediction of the outcome. However, it is nearly impossible to predict outcome based on first impressions and Apgar scores, there is no evidence that first impressions can be used to postpone the decision whether to initiate life sustaining treatment or not till the baby is born.[14] Parental hope on the usefulness of first impressions should be refuted. Furthermore, it is importance to address uncertainties and the impossibility to predict an individual outcome.[15,16] However, doctors should share what they know to provide a more individualized prediction of the outcome. For example, parents could be better guided in the process of decision-making by informing them about how various factors could influence neonatal outcomes and by providing them with information on postnatal predictors of prognosis, including any potential treatment limitations.[16-18]

Secondly, participants wished the impact of extremely premature birth on the entire family to be considered in prenatal decision-making, particularly on siblings and the financial consequences for the family. Research has shown that the effects of a premature infant with a disability can have substantial impact on the family[11, 19, 20], especially the negative consequences of reduced parental attention for siblings[11, 21], and feelings of loyalty and responsibility toward the prematurely born infant.[21] By discussing the impact of premature birth on families, parents can incorporate this into their decision-making process and determine whether additional support for the family is needed.

Lastly, participants emphasized the importance of informing parents more about the long-term consequences of extremely premature birth. While the consequences during childhood are widely recognized by healthcare professionals and parents, our participants emphasized the lifelong consequences beyond childhood, as well as the lack of aftercare later in life. Research on the consequences and outcomes of adults who were born extremely prematurely is increasingly available.[10,22-28] Studies demonstrate a higher risk for poor social outcomes in adulthood[24,25,29] and an increased risk for long-term morbidities.[27] Participants showed that in their individual cases, their parents felt unprepared for these long term issues. It is unknown why neonatologists do not put emphasis on functional long-term outcomes in prenatal counselling. It may be that they are not sufficiently aware of these long-term consequences, or that they do not consider it a priority topic, or maybe parents do not consider it important enough and consequently focus on the present reality.[5] Participants want future parents to be informed about the challenges their infant may encounter in healthcare, school, work and personal life, which may continue or arise beyond childhood.[10,27,28]

However, presenting data on long-term outcomes in adulthood comes with an inherent dilemma for HCPs, as data is inherently collected ages ago. Changes in patient care have occurred in the evolving field of neonatology.[30] Treatments that are given today may have positive or negative influence on long-term consequences and those can only be known through long-term follow-up. It is important to acknowledge this complexity while counselling parents about the long-term outcomes beyond childhood. Nevertheless, prenatal treatment decisions on resuscitation must to be made based on the best available information. Furthermore, our results merely show that the impact on a premature born person and his family is lifelong and even though emphasis may switch over time from one medical or psychological problem to another, the main message is to discuss the fact that the impact is lifelong and that is unlikely to change.

Therefore, it is essential to (I) discuss the potential lifelong consequences and challenges in various aspects of life while acknowledging that the information is derived from ‘older' but only available data, and (II) make parents aware of the lack of follow-up beyond early childhood.[31]

Strengths and limitations

The major strength of this study is the uniqueness of participants; the prematurely born adults. However, our study also has some limitations. Firstly, the major limitation is the possible survivor bias caused by the inherent non-representative sample of participants. Obviously, only those for whom an EIC-treatment was chosen and survived could be included. Participants were only included when they had the intellectual and communicative level to be able to participate. Furthermore, it is uncertain how the heterogeneity in individuals' outcomes with regards to the level of disabilities influences their perspective (whether positive or negative) on these outcomes. Consequently, the composition of the sample also affects the results. However, our participants experienced various consequences of their extremely premature birth, like cerebral palsy or BPD. It is impossible to include non-survivors and adults with severe cognitive impairments. Additionally, recruitment through social media and the patient organization could have caused a selection bias.

A second limitation is that the study focused on participants from the Netherlands where the standard of neonatal care and societal and cultural values may differ from those in other countries. The Dutch context prioritizes quality of life[32], which may have influenced the perspectives of the participants. Furthermore, it is a high-income country which influences the availability of neonatal intensive care.

A third limitation may be that our results are limited by reporter bias, due to the study design of online focus group discussions. This design may not always allow for a full expression of thoughts or feelings, and may have affected the group dynamics although unknown in which way. However, during the individual meetings afterwards, none of the participants reported any issues related to these limitations.

Lastly, recall bias could have limited the results by the potential misrepresentation or inaccuracy in participants' recollection of past events or experiences. However, we explored their perspective on what they now consider important based on their experiences in life and did not ask about one particular event in the past.

Conclusion

To the best of our knowledge, this is the first qualitative study with extremely prematurely born adults on what they consider important values in prenatal decision-making at the limit of viability. Their perspectives have so far been underrepresented but could contribute to improving care for (future) parents and their infants. Our study suggests that it is crucial to address the expectations and uncertainty regarding individual prognosis.[17] Additionally, the results emphasize the importance of providing parents with comprehensive information about potential consequences for both their infant and their family associated with an extremely premature birth. Extremely premature birth should be considered to be a lifelong diagnosis.[25]

References

1. Lemyre B, Daboval T, Dunn S, Kekewich M, Jones G, Wang D, et al. Shared decision making for infants born at the threshold of viability: a prognosis-based guideline. J Perinatol. 2016;36(7):503-9.
2. Kaempf JW, Tomlinson MW, Tuohey J. Extremely premature birth and the choice of neonatal intensive care versus palliative comfort care: an 18-year single-center experience. J Perinatol. 2016;36(3):190-5.
3. Kukora SK, Boss RD. Values-based shared decision-making in the antenatal period. Semin Fetal Neonatal Med. 2018;23(1):17-24.
4. Stiggelbout AM, Pieterse AH, De Haes JC. Shared decision making: Concepts, evidence, and practice. Patient Educ Couns. 2015;98(10):1172-9.
5. de Boer A, de Vries M, Berken DJ, van Dam H, Verweij EJ, Hogeveen M, et al. A scoping review of parental values during prenatal decisions about treatment options after extremely premature birth. Acta Paediatr. 2023.
6. Moro TT, Kavanaugh K, Savage TA, Reyes MR, Kimura RE, Bhat R. Parent decision making for life support for extremely premature infants: from the prenatal through end-of-life period. J Perinat Neonatal Nurs. 2011;25(1):52-60.
7. Hendriks MJ, Abraham A. End-of-Life Decision Making for Parents of Extremely Preterm Infants. J Obstet Gynecol Neonatal Nurs. 2017;46(5):727-36.
8. Tucker Edmonds B, Hoffman SM, Laitano T, Jeffries E, Jager S, Kavanaugh K. Diverse perspectives on death, disability, and quality of life: an exploratory study of racial differences in periviable decision-making. J Perinatol. 2021;41(3):396-403.
9. Lynch TA, Cheyney M, Chan M, Walia J, Burcher P. Temporal Themes in Periviable Birth: A Qualitative Analysis of Patient Experiences. Matern Child Health J. 2019;23(3):422-30.
10. Saigal S. In their own words: Life at adulthood after very premature birth. Semin Perinatol. 2016;40(8):578-83.
11. Saigal S, Burrows E, Stoskopf BL, Rosenbaum PL, Streiner D. Impact of extreme prematurity on families of adolescent children. J Pediatr. 2000;137(5):701-6.
12. Vohr BR. How should we report early childhood outcomes of very low birth weight infants? Semin Fetal Neonatal Med. 2007;12(5):355-62.
13. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. Int J Qual Health Care. 2007;19(6):349-57.
14. Singh J, Fanaroff J, Andrews B, Caldarelli L, Lagatta J, Plesha-Troyke S, et al. Resuscitation in the “gray zone” of viability: determining physician preferences and predicting infant outcomes. Pediatrics. 2007;120(3):519-26.
15. Prins S, Linn AJ, van Kaam A, van de Loo M, van Woensel JBM, van Heerde M, et al. How Physicians Discuss Uncertainty With Parents in Intensive Care Units. Pediatrics. 2022;149(6).
16. Geurtzen R, van Heijst AFJ, Draaisma JMT, Kuijpers L, Woiski M, Scheepers HCJ, et al. Development of Nationwide Recommendations to Support Prenatal Counseling in Extreme Prematurity. Pediatrics. 2019;143(6).
17. Wilkinson AR, Ahluwalia J, Cole A, Crawford D, Fyle J, Gordon A, et al. Management of babies born extremely preterm at less than 26 weeks of gestation: a framework for clinical practice at the time of birth. Arch Dis Child Fetal Neonatal Ed. 2009;94(1):F2-5.
18. Wood K, Di Stefano LM, Mactier H, Bates SE, Wilkinson D. Individualised decision making: interpretation of risk for extremely preterm infants-a survey of UK neonatal professionals. Arch Dis Child Fetal Neonatal Ed. 2022;107(3):281-8.
19. O’Donovan A, Nixon E. “Weathering the storm:” Mothers’ and fathers’ experiences of parenting a preterm infant. Infant Ment Health J. 2019;40(4):573-87.
20. Treyvaud K, Lee KJ, Doyle LW, Anderson PJ. Very preterm birth influences parental mental health and family outcomes seven years after birth. J Pediatr. 2014;164(3):515-21.
21. Gaal BJ, Pinelli J, Crooks D, Saigal S, Streiner DL, Boyle M. Outside looking in: the lived experience of adults with prematurely born siblings. Qual Health Res. 2010;20(11):1532-45.

22. Cheong JLY, Haikerwal A, Anderson PJ, Doyle LW. Outcomes into adulthood of infants born extremely preterm. Semin Perinatol. 2021;45(8):151483.

23. Moster D, Lie RT, Markestad T. Long-term medical and social consequences of preterm birth. N Engl J Med. 2008;359(3):262-73.

24. Day KL, Van Lieshout RJ, Vaillancourt T, Saigal S, Boyle MH, Schmidt LA. Long-term effects of peer victimization on social outcomes through the fourth decade of life in individuals born at normal or extremely low birthweight. Br J Dev Psychol. 2017;35(3):334-48.

25. Hille ET, Dorrepaal C, Perenboom R, Gravenhorst JB, Brand R, Verloove-Vanhorick SP. Social lifestyle, risk-taking behavior, and psychopathology in young adults born very preterm or with a very low birthweight. J Pediatr. 2008;152(6):793-800, .e1-4.

26. Pyhälä R, Wolford E, Kautiainen H, Andersson S, Bartmann P, Baumann N, et al. Self-Reported Mental Health Problems Among Adults Born Preterm: A Meta-analysis. Pediatrics. 2017;139(4).

27. Raju TNK, Buist AS, Blaisdell CJ, Moxey-Mims M, Saigal S. Adults born preterm: a review of general health and system-specific outcomes. Acta Paediatr. 2017;106(9):1409-37.

28. Saigal S. Preemie Voices: Young men and women born very prematurely describe their lives, challenges and achievements: FriesenPress; 2014.

29. Saigal S, Day KL, Van Lieshout RJ, Schmidt LA, Morrison KM, Boyle MH. Health, Wealth, Social Integration, and Sexuality of Extremely Low-Birth-Weight Prematurely Born Adults in the Fourth Decade of Life. JAMA Pediatr. 2016;170(7):678-86.

30. Philip AG. The evolution of neonatology. Pediatr Res. 2005;58(4):799-815.

31. Perez A, Thiede L, Lüdecke D, Ebenebe CU, von dem Knesebeck O, Singer D. Lost in Transition: Health Care Experiences of Adults Born Very Preterm-A Qualitative Approach. Front Public Health. 2020;8:605149.

32. De Proost L, Verweij EJT, Ismaili M'hamdi H, Reiss IKM, Steegers EAP, Geurtzen R, et al. The Edge of Perinatal Viability: Understanding the Dutch Position. Front Pediatr. 2021;9:634290.

Appendix 1: interviewguide TINY-1

1. *A short introduction for the focus group*

   In short: welcoming the participants, explaining the aim of the research and the rules. Followed by a round in which the participants could introduce themselves and why they were participating.

2. *Introduction of the subject*

   In short: explaining a normal pregnancy is 40 weeks. Followed by an explanation of the grey zone, the treatment options at the limit of viability, and the decision that has to be made together with parents. Part of this decision is to elaborate what they think is important in life.

3. *Part I: Periviable guidelines & Part II: Personalization of care (not the scope of this thesis)*

4. *Part III: values*

   In this part of the focus group we want to discuss what you think is important in decision-making when extremely premature birth threatens. When there is no best choice, because advantages and disadvantages are influenced by what you/your parents/your family consider important in life, it is recommended to decide together with parents what decision is best. In such a decision, various aspects or arguments can play a role for parents and families. You can also call these values.

a) Values important in decision-making
- What arguments/values do you think should be important in making a decision regarding treatment after birth at the limits of viability?
  - o Exploring the named arguments/values/perspectives with questions like
    - ● What do you mean when you say [value/argument/consideration]?
    - ● If you say [value/argument/consideration], what should we be considering? Etc.

b) We searched the existing literature on what parents consider important in making a decision whether to initiate intensive care or comfort care after premature birth. The results we found are displayed in a figure. We are going to show the figure (*see chapter 2)* The size of the circles is based on the number of articles in which the perspective is mentioned.
- What comes to mind when seeing this figure?
- Which perspectives in the figure do you recognize, or you do not recognize? And why?

Appendix 2: codebook

Theme	Subcode	Description
Financial impact	Financially barrable?	Considering in decision-making if it is financially bearable for the family?
	Financial support parents	Considering in decision-making if parents could financially support a possible disabled child with its financially consequences
	Financially stable situation	Considering in decision-making if parents have a financially stable situation
Got lucky	Aware of miracle	Be aware of the miracle to be born healthy or not suffering from serious consequences
	Thankful for life	Thankful for the life or being a life
	Way of life	Being lucky, in context of how you live your life as a result of the knowledge you got lucky
	How it could have been	How it also could have been, for example serious disabilities
	Few consequences	Having few till no consequences of the premature birth
Give a chance	Chance of survival	Choosing intensive care treatment when there is a chance of survival
	End of life decision in the NICU	Choosing intensive care treatment with the knowledge the decision to end life support in the NICU is possible when treatment is not proportional anymore
	Try/everything done/save	Choosing intensive care to try and do everything
Impact on family-system	Siblings	Take the impact on the siblings into account
	Context of family	Consider the context of the family
	Parental capacity	Important to judge the parental capacity, otherwise extra help is needed
	Negative	Someone mentioning this should not be considered
Individualize	Every decision is a good one	You cannot make a bad decision
	Value clarification each parent	Value clarification should be performed, with each parent on its own
	Values individual process	Forming values and having values are an individual process
	Values differ per individual	Everybody has his own values
	Which knowledge/numbers are needed/wanted	Individualize the knowledge you give, the number you mention based on what a individual wants to hear
Intuition	Could go wrong	Making a decision based on intuition or fully emotional, could go wrong
	More intuition than deliberation of values	Could imagine intuition plays a bigger part in decision-making, than deliberation of values

Theme	Subcode	Description
	Feeling of a mother/ instinct	It is just a feeling of a mother, a primal instinct
	Peace	Peace is necessarily
Look at the child	Survival instinct	Making the decision based on the instinct to survive the child shows
	Situation infant post-partum	Making the decision based on what the child shows after birth
	Fighters	Making the decision based on if the child shows it wants to fight for life
	Examples of factors	Examples of factors that should be looked at after birth to estimate the will to live
Protect against burden of short- and long-term outcomes (LTO)	Short term outcomes	Mentioning short term outcomes to consider (NICU-treatment)
	LTO - possible suffering	Considering the possible suffering for the child resulting from long-term consequences
	LTO – not an easy life	Considering long-term outcomes which could lead to a difficult life
	LTO – consequences for the child	Considering consequences for the child
	LTO – 'kasplantje' (=dutch expression meaning vegetable)	Considering the prediction of being a 'kasplantje'
	LTO – difficult to predict	Long-term outcomes are difficult to predict
	LTO – future research	Future research has to be done in this field
	LTO – consequences for the adults	Considering consequences for adults who were born prematurely
	LTO – wrong prediction	Be careful with predicting outcomes because it could be wrong
	LTO – educate parents	Inform parents about the possibilities of long-term consequences
	LTO – what is acceptable	What are acceptable outcomes? Differs between individuals?
Quality of life	Context of a healthy life	Quality of life in the context of a healthy life
	How to determine	Quality of life is important, but how do you determine what is quality of life and who is going to do that
	Individual	It differs per individual what quality of life is
	Quality of life	Only quality of life was mentioned as important
	Pleasure in life	The possibility of pleasure in life should be considered as part of quality of life in decision-making

Theme	Subcode	Description
	Unpredictable	Very hard to predict if a child would have a good quality of life, but it still should be considered with the available knowledge
	Prediction	Predicting quality of life with the available knowledge
Quote	(Theme to use with)	Quote
Taking control	Include in decision-making	Parents have to be included in decision-making
	To take control	Considered important that parents have control in decision-making
	To take away control	Be aware of things that could take control away from parents, like not clarifying values
	Which part in decision-making	Important to include parents in decision-making, but always ask which part they want to play
Religion	No active termination of life	Religions could say active termination of life is not acceptable
	Values formed out of religion	Could be imagined that values based on religion could be important for some parents
Anticipated guilt/regret	Possible suffering	The consideration of anticipated guilt about the suffering of a child when faced with complications after birth
	Wrong choice	The consideration of anticipated guilt when making the wrong choice, either intensive care or comfort care
	Different perspective of the child	Different views of parents and the child related to the possibility of regret or guilt
SDM	Afraid to make the wrong choice	Being part of the decision, could make parents afraid to make the wrong decision
	Uncertainty	Making a decision together while there is so much uncertainty
	Hardest decision	It is the hardest decision there is
	Process of decision-making/transparency	Include parents in the process of decision-making, be transparent
Social support	Social support	Considering social support in the decision-making
	Support from environment	Considering the social environment of parents in the decision-making
Spiritualty	It has to be like this	To be alive, it has to be like this
	Connection with the child	Estimate which decision you have to make based on the connection with your child
Survival	Hope to survive	Hope on survival
	Chance of survival	The chances of survival
	Despite outcomes/consequences	Survival at all, despite which outcomes/consequence there are, it is all acceptable
Responsibility	Advocate for the child	Feeling responsible for their child and therefore fighting/advocating for them