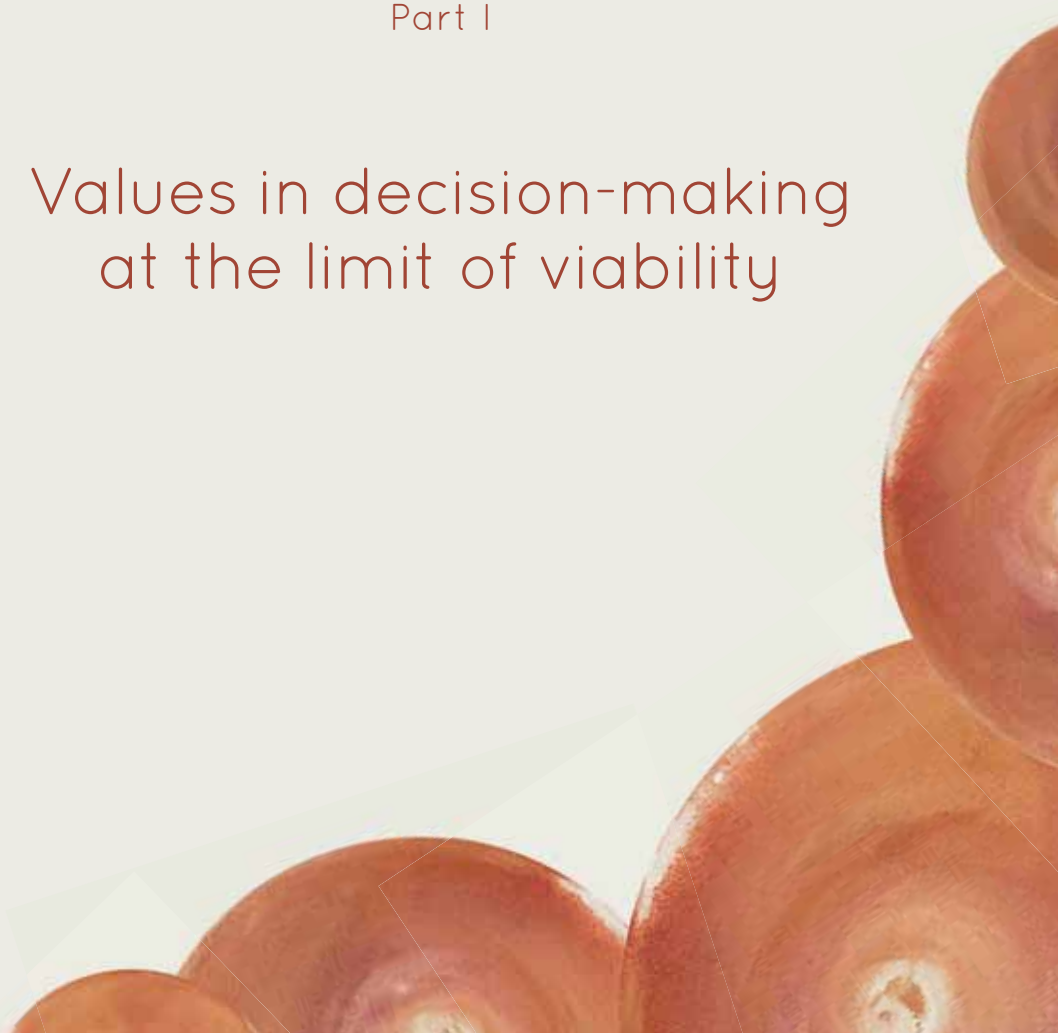
Voices of experience in periviable decision-making and artificial placenta technology

Boer, A.H.A. de

Part I

Values in decision-making at the limit of viability



Chapter 2

A scoping review of parental values during prenatal decisions about treatment options after extremely premature birth

Angret de Boer, Marieke de Vries, Dirk-Jan Berken en Hiske van Dam,
E.J.T. (Joanne) Verweij, Marije Hogeveen, Rosa Geurtzen.

Acta Paediatrica, 2023

Abstract

Aim: To describe what is known in the literature about parental perspectives in making prenatal decisions about treatment after birth at the limit of viability, as a better understanding of parental values can help professionals support parents as they decide.

Methods: PubMed, Cochrane, Embase, CINAHL, PsycINFO and Web of Science were searched to identify relevant literature from 1 January 2010 to 22 April 2022 on parental decision making. Data were extracted from selected studies and organized into themes. The final themes were formed through collaboration with the parents of a premature infant born at 24 weeks.

Results: Of the 15,159 papers examined, 17 were included. Parental perspectives were described in terms of long-term outcomes for the infant, survival, protection against the burden of neonatal treatment, long-term impact on the family, religion and spiritual beliefs, to do everything possible, hope, sense of responsibility, wanting the best, doing what is right, giving a chance and the influence of experience.

Conclusion: The extracted parental perspectives show the complexity of these decisions. Some perspectives were clear, but others were multi-interpretable. Increasing the understanding of common parental perspectives can help improve shared prenatal decisions and lead to further improvement and personalisation of the process**.**

Background

Parents need to be involved in prenatal decisions about neonatal treatment if their baby is likely to be born extremely preterm -- in the grey zone, which is between 22 and 26 weeks of gestation, depending on time, country and culture.[1-3] The two treatment options in this grey zone are intensive care and comfort care. Intensive care treatment comes with great uncertainty regarding the outcome: some infants will not survive, some will go through life with one or more disabilities, and others will grow up without problems.[4]

As decisions in the grey zone are value laden,[5] a shared decision-making approach is recommended.[6-9] This involves mutual recognition of the need for a decision, followed by a discussion of the treatment options with their pros and cons, as well as an exploration of the parental values to be incorporated.[10,11] This exploration, known as *values clarification*, will allow treatment decisions to be aligned with personal goals and circumstances.[12] When a prenatal decision must be made, the preferences and values of the parents, who represent the unborn child, should be explored.[13]

Previous research has focused mainly on decision-making models and the preferred roles of parents and healthcare providers as they make decisions.[14] Although clarifying values is considered an essential aspect of shared decision making,[8,15,16] it is not always practised in prenatal decision making.[16] Improved understanding and identification of common parental values can help professionals support parents as they decide. The aim of this study, therefore, is to describe what has been published in the literature about the parental perspectives involved in prenatal decision making in cases of extreme prematurity.

Methods

This scoping review was guided by an article written by Micah Peters[17] and the checklist provided in the PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation; PRISMA stands for Preferred Reporting Items for Systematic reviews and Meta-Analyses.[18]

Search and study selection

In consultation with a literature specialist, we systematically searched PubMed, Cochrane, Embase, CINAHL, PsycINFO and Web of Science. The complete PubMed search is provided in *Appendix S1*. The databases were searched for literature published between 1 January 2010 and 22 April 2022. Reference lists of the included papers and related systematic reviews were screened to identify additional references. The results were collected in Endnote X9, a reference management tool (Clarivate, Philadelphia, USA) and deduplicated prior to screening. The deduplicated results were imported into an online reviewing system (Rayyan, Oxford, UK).[19]

Two authors (AB and RG) independently performed a title and abstract screening, followed by a full-text screening. The eligibility criteria are summarised in *Table 1*. Disagreements were settled through discussion with two authors (MV and MH).

Table 1: Eligibility criteria

Inclusion criteria	Exclusion criteria
The focus of the paper on prenatal decision-making regarding treatment after birth at the limit of viability; intensive care treatment or comfort care	Does not meet the inclusion criteria
The research population involves parents who experienced actual or imminent extremely premature birth at the limit of viability	Guidelines, expert opinions and reviews
Describing parental perspectives, arguments, considerations or specific values regarding the treatment options at the limit of viability	Publication that reported prompted perspectives, so they were not spontaneously named by the parents
English or Dutch	Unable to retrieve the abstract or the full-text paper
	Publications performed before 2010

Data analysis

Data pertaining to parental perspectives were charted from the included studies, using a data extraction tool made in Microsoft Excel (Microsoft 365 MSO, version 2202, UK).[17] We have used the term parental perspectives in this article to refer to several aligned items, such as parental perspectives, considerations, arguments, values and views. The key findings reported in *Table S2* are those of our research aim and are not necessarily similar to the key findings of the original manuscripts.

The authors developed a coding strategy prior to data extraction and adjusted it throughout the review process. To minimise extraction errors, two authors (AB and RG) independently extracted text parts from the included manuscripts. When papers did not exclusively focus on extreme prematurity or parents, the potentially eligible data were extracted except when they were explicitly described in another context. Thematic analysis was used to organise the extracted data into themes, which were substantiated with illustrative quotes from the included studies. Our research team (AB, RG, MV, MH and JV) collaborated with the parents (HD and DB) of a premature infant born at 24 weeks in 2020 to discuss and form the final themes derived from the data.

Results

Figure 1 presents the PRISMA flowchart of the inclusion process. The literature search revealed 15,159 deduplicated records, from which 17 papers were selected for inclusion in this review. Most of the studies were conducted in the United States (n=12)[20-31] and the others came from Canada (n=2),[32,33] Switzerland (n=1),[34] Norway (n=1)[35] and the Netherlands (n=1).[14] An overview of the characteristics of all included studies can be found in *Table*

S2. The results are charted in order of frequency, starting with the theme that was covered in the most papers. *Figure 2* provides a graphical display of the results.

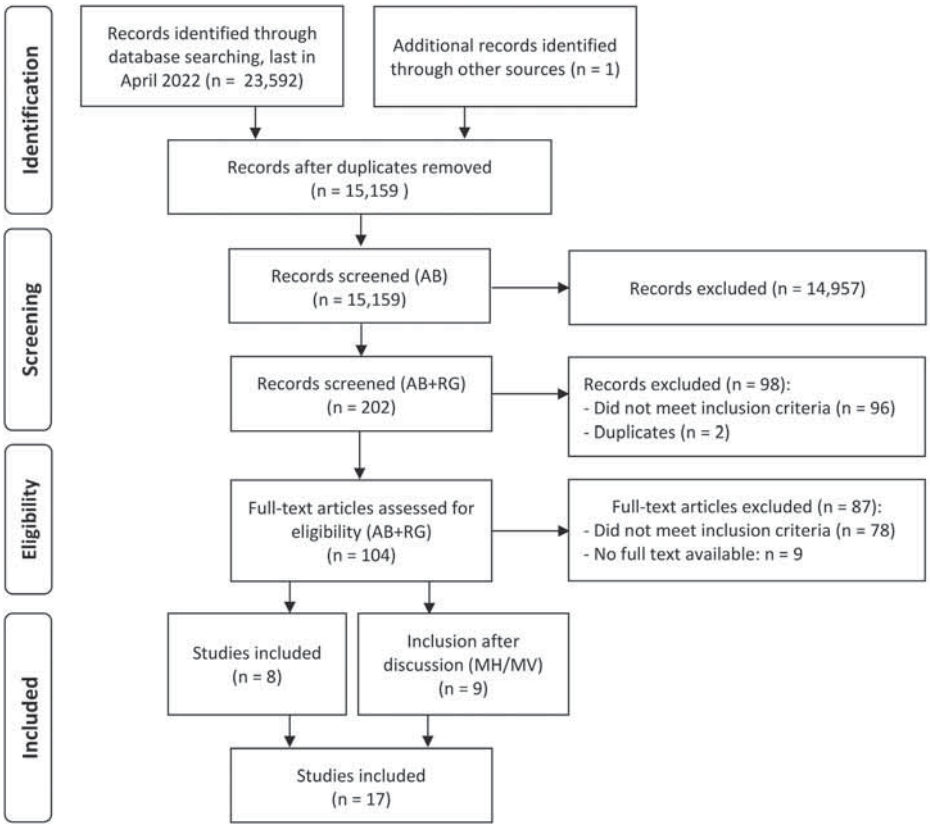


Figure 1 Prisma flowchart of results

Long-term outcomes for the infant

Many parents featured in the papers were concerned about the potential long-term outcomes for their premature infant, in terms of disabilities,[14,20,22,24,28,29,33,35] neurological or non-neurological complications[14,25,29,30,32,33] and quality of life.[14,27,29,30] These concerns were often cited as factors in parental decision-making about neonatal treatment, with some parents opting for comfort care. However, many parents chose intensive care despite these concerns, as they prioritised the infant's life. Opinions about acceptable long-term outcomes and the perception of the presumed quality of life of a disabled child were personal and differed amongst parents, as did the extent to which those outcomes played a role in the decision. As one parent stated: "*If I deliver this early, knowing that baby could have -- you know -- disabilities is the reason why I said do not resuscitate*".[24] Other parents said they may not have chosen resuscitation if they had known the potential complications[32] or to protect their child from becoming a vegetable.[30]

Other long-term considerations and preferred outcomes were described as a dignified existence[14] or a healthy infant[24], leading either to a decision for comfort care or an unspecified decision. Independence, participation in society and prognosis of intact survival were also reported as important considerations.[14]

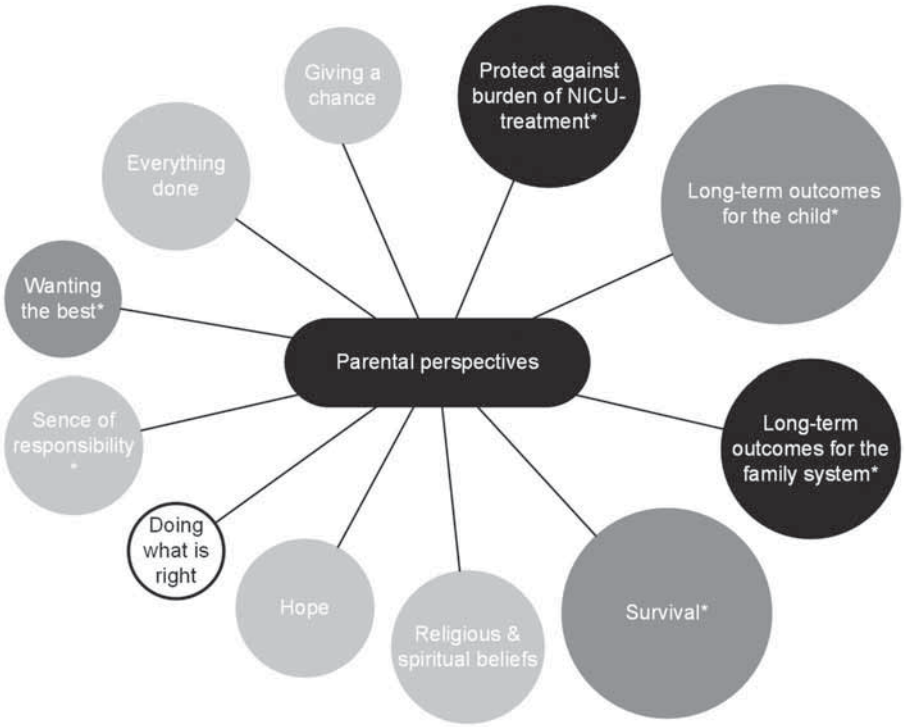


Figure 2 Graphical display of the parental perspectives extracted from the included literature. Each perspective is circled, with the circle radius proportional to the number of papers in which it is mentioned. The colour of the circle is based upon the choice (if this was recorded) made by the parents who expressed this perspective. Black = comfort care, light grey = intensive care treatment, dark grey = both comfort care and intensive care treatment. A white circle or a * in the circle means that in some papers in which parents expressed this perspective, no mention was made of the parental choice.

Parental concerns regarding the infant's future quality of life were also raised. One parent said, "*So, I think the majority of that decision would be based on the quality of life that your child would have".*[30] These concerns led to decisions for comfort care or for decisions that remained unspecified.

Survival

This theme covered different aspects of survival, including its general potential and specific probability. Some parents viewed the potential for survival as a reason to initiate intensive care treatment, while others, having weighed the probability of survival against the risk of short-term suffering or complications, opted for comfort care. When parents believed there

was a reasonable chance of long-term survival, they were likelier to choose intensive care. However, some parents were not concerned about potential complications and morbidities as long as their child survived.[14,20,26,29-31] Others were more focused on avoiding unnecessary suffering for their infant if the chances of survival were low[27,33]; as one parent argued: "*We don't want to have to put a child through something that she's not going to survive anyway*".[27]

Protection against the burden of treatment

When faced with the decision, parents were concerned about the pain and suffering that intensive care treatment could cause.[14,24,27,29-32,34] They weighed up the disadvantages of potential suffering against the benefits of comfort care.[27] One mother argued that the benefit of not resuscitating was "*that the baby would not be in pain and would probably pass quickly and not with stress*".[27] It was reported that parents would not have initiated intensive care treatment had they known how much their child would have to suffer in the neonatal intensive care unit (NICU).[32] However, although most parents considered short-term suffering to be an important argument in favour of comfort care, some still chose intensive care treatment in spite of their concerns about it.

Long-term impact on the family

Some parents reported concerns about long-term outcomes for themselves and the impact on other family members when choosing intensive care treatment.[14,26,28,29,32,35] One study reported parents as saying that family life, which also matters, could be very different with a premature child.[35] In two papers based on data gathered in the USA, mothers expressed their fear of the financial consequences.[23,25]

Religion and spiritual beliefs

Several papers discussed parents' reliance on spirituality, faith and religion to make the decision to initiate intensive care treatment.[20,21,24,27,29,30,33] Parents believed the decision should be left to "*God*".[20,21] Faith helped them to maintain hope[20] and not give up, despite the risks.[27] One mother argued: "*She's gonna be fine ... I keep referring back to God has the last say, so ... He's not gonna give me or my partner too much that we can't bear*".[30]

Everything done

In some of the papers, parents justified their decision for intensive care treatment by saying they wanted everything done.[14,26,30,31,34,35] As one parent explained, "*We had the option not to do anything and then she would die after a while. Or we had the option to try everything possible ... We discussed this and we said we would try everything*".[34] The authors of one study worried that failure to define the concept of everything done could cause a misperception of the delivered care. One of the parents in that study felt that her infant did not receive appropriate care in spite of her insistence that everything should be done.[31]

Hope

Hope appeared to be important in prenatal counselling, especially for parents who chose intensive care.[20,27,29,31,35] Despite the physicians' medical information about risks of disability or death, parents based their decision on their hopes that their infant would survive, would be fine or would be better than expected.[31] One article reported that some parents wanted hope while others wanted realism.[29] Hope could be helpful for parents. As one stated: "*The pain is going to be there no matter what, so I didn't mind trying to have hope".*[29] This theme had some overlap with the Survival subtheme described above.

Sense of responsibility

Papers addressing this theme reported that parents chose either intensive care treatment or made no clear decision. Several papers described how parents felt they had to advocate for their infant and take responsibility.[14,24,25,35] Parents felt responsible for making the final decision[14,30,35] or were convinced that it was their job to take responsibility for and protect their infant.[14] They needed to be assured that there was no right or wrong in making their decision.[14] Another article described how parents experienced an instinct to save the infant; as one explained: "*You experience that the survival instinct when you are pregnant, and the protection instinct after birth, is so strong that it is difficult in the situation … I do not know how long it took me to arrive at a more nuanced view".*[35]

Giving a chance

Parents choosing intensive care treatment described giving the infant a fighting chance as important.[14,24,27,30] One mother justified her decision by saying: "*Give her an opportunity, give her a chance, don't write her off".*[27] Choosing comfort care felt like denying the infant the chance to fight,[14] and parents just wanted to give their infant a chance to live because they felt the infant deserved it as much as any other baby.[27]

Wanting the best for the infant

In some of the studies, parents argued that they wanted the best for their infant[24,26,35] and act in the child's best interest[35]: "*The whole thought process the whole time was … just whatever is going to be the best outcome for her".*[26] However, parents differed in their interpretations of the best; for some, it meant choosing intensive care treatment, for others, comfort care.

Doing what is right

While making their decision, parents wanted to do what was most '*natural and right'*, without specifying to what natural and right pertained or to which decision this led.[20,22] Another article reported parents as saying they anticipated their possible regret.[29]

Influence of experience

Parents' decisions could be influenced by prior experiences, either their own[20,24] or an acquaintance's,[21,24,29] with a premature or extremely premature infant treated in the NICU.

Discussion

We carried out a scoping review of parental perspectives on decision making at the limits of viability. Our results offer insight into the most common values underlying parental decisions. Complex and multi-layered, some themes reflect the weighing of factual information about outcomes for infants and families, as well as numerous underlying and potentially conflicting values. Other themes reflect a process preference, such as the desire to do everything possible, or reflect feelings or intuitions, such as the instinct to save the child. The results demonstrate parents' difficulties in clarifying and verbalising their values in a sudden situation that provokes anxiety and is laden with values. Similarly, physicians may not be fully equipped to clarify parental values and fail to recognise that parental perspectives can be unclear and open to interpretation or misinterpretation.[15,36-38] This can make it challenging to provide guidance and support to parents. One perspective can be interpreted differently by different parents and lead to different decisions, so that some will decide to initiate intensive care treatment and others comfort care, based on the same perspective. For example, some parents could believe that everything done means everything technically possible, while others think the phrase means that they simply want to feel that they have done all they could for their child.[39]

The results of this review raise questions about the extent to which feelings and intuitions, rather than explicit deliberations about values, guide the choice of intensive care treatment or comfort care. Themes, such as everything done or hope, suggest that feelings or intuitions may be more important than explicit deliberations about values. However, the extent to which these feelings are based on underlying values is unclear because these values, difficult as they may be to verbalise explicitly, may be genuinely important to parents in the long run.[40] If this is the case, these feelings may help guide the making of decisions in this context, which is highly sensitive to preferences. However, it can be challenging to distinguish biased feelings or intuitions from those that tap into genuine underlying parental values. Feelings may arise in the heat of the moment, be based on perceived social norms or coloured by information from a biased counsellor; such feelings are not good bases for decisions in the long run because they can overshadow other important aspects. More research is needed not only to understand how much reliance parents place on intuition or thought when coming to a decision, but also whether it is wrong to incorporate emotion in this decision.[40]

To navigate these challenges in clinical practice, parents could be allowed a little extra time. This would allow them to process the information and the overwhelming emotions arising from their first reactions, thereby developing a more balanced feeling. The delay may help them consider all relevant aspects of the decision rather than just reacting to the initial shock or stress.[41] Even under the time pressure inherent in (imminent) extreme premature birth, a little extra time may help parents to make decisions that align with their values. Another suggestion is to allow parents to talk to a significant other.[42] However,

it is not always feasible for parents to fully process and understand the situation before making a decision, so it is important for healthcare professionals to continue supporting and checking in with parents postnatally to ensure that decisions are made with care and consideration.[15]

Limitations

This scoping review had some limitations. First, the depth of our results could be limited because we extracted data from the summarised documents rather than from the original interview transcripts. Second, it was impossible to count the exact number of parents mentioning each theme or to systematically compare representations of perspectives between the groups opting for intensive care and comfort care because the raw data was lacking. Third, we may have missed some grey literature outside the healthcare databases that might have included parental perspectives. Fourth, the assembled data might have been biased; cultural bias might have been present because most of the studies were conducted in the USA, or a bias might have existed towards intensive care treatment because most parents featured in the selected literature either chose intensive care treatment or made no clear, recorded decision. Finally, the widespread timeframe of parent interviews -- some were interviewed before the premature birth, some years afterwards -- may have had an impact on the perspectives parents considered important.

Conclusion

This review highlights common perspectives that affect parents' decisions at the limit of viability. An increased understanding of parental perspectives and their underlying values can improve the clarification values as part of shared decision making. Barriers should be minimised and facilitators leveraged to promote and improve the clarification of values in prenatal counselling. This may improve prenatal counselling and promote value-congruent decisions, a key goal in patient-centred care.[24]

References

1. Gallagher K, Martin J, Keller M, Marlow N. European variation in decision-making and parental involvement during preterm birth. Arch Dis Child Fetal Neonatal Ed. May 2014;99(3):F245-9. doi:10.1136/archdischild-2013-305191
2. Guillén Ú, Weiss EM, Munson D, et al. Guidelines for the Management of Extremely Premature Deliveries: A Systematic Review. Pediatrics. Aug 2015;136(2):343-50. doi:10.1542/peds.2015-0542
3. Gillam L, Wilkinson D, Xafis V, Isaacs D. Decision-making at the borderline of viability: Who should decide and on what basis? J Paediatr Child Health. Feb 2017;53(2):105-111. doi:10.1111/jpc.13423
4. Myrhaug HT, Brurberg KG, Hov L, Markestad T. Survival and Impairment of Extremely Premature Infants: A Meta-analysis. Pediatrics. Feb 2019;143(2)doi:10.1542/peds.2018-0933
5. 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. Mar 2016;36(3):190-5. doi:10.1038/jp.2015.171
6. Geurtzen R, van Heijst AFJ, Draaisma JMT, et al. Development of Nationwide Recommendations to Support Prenatal Counseling in Extreme Prematurity. Pediatrics. Jun 2019;143(6)doi:10.1542/peds.2018-3253
7. Cummings J, Committee On F, Newborn. Antenatal Counseling Regarding Resuscitation and Intensive Care Before 25 Weeks of Gestation. Pediatrics. Sep 2015;136(3):588-95. doi:10.1542/peds.2015-2336
8. Kukora SK, Boss RD. Values-based shared decision-making in the antenatal period. Semin Fetal Neonatal Med. Feb 2018;23(1):17-24. doi:10.1016/j.siny.2017.09.003
9. Mactier H, Bates SE, Johnston T, et al. Perinatal management of extreme preterm birth before 27 weeks of gestation: a framework for practice. Arch Dis Child Fetal Neonatal Ed. May 2020;105(3):232-239. doi:10.1136/archdischild-2019-318402
10. Stiggelbout AM, Pieterse AH, De Haes JC. Shared decision making: Concepts, evidence, and practice. Patient Educ Couns. Oct 2015;98(10):1172-9. doi:10.1016/j.pec.2015.06.022
11. Elwyn G, Frosch D, Thomson R, et al. Shared decision making: a model for clinical practice. J Gen Intern Med. Oct 2012;27(10):1361-7. doi:10.1007/s11606-012-2077-6
12. Fagerlin A, Pignone M, Abhyankar P, et al. Clarifying values: an updated review. BMC Med Inform Decis Mak. 2013;13 Suppl 2(Suppl 2):S8. doi:10.1186/1472-6947-13-s2-s8
13. Barker C, Dunn S, Moore GP, Reszel J, Lemyre B, Daboval T. Shared decision making during antenatal counselling for anticipated extremely preterm birth. Paediatr Child Health. Jul 2019;24(4):240-249. doi:10.1093/pch/pxy158
14. Geurtzen R, van Heijst A, Draaisma J, et al. Prenatal counseling in extreme prematurity - Insight into preferences from experienced parents. Patient Educ Couns. Aug 2019;102(8):1541-1549. doi:10.1016/j.pec.2019.03.016
15. Haward MF, Payot A, Feudtner C, Janvier A. Personalized communication with parents of children born at less than 25 weeks: Moving from doctor-driven to parent-personalized discussions. Semin Perinatol. Mar 2022;46(2):151551. doi:10.1016/j.semperi.2021.151551
16. Tucker Edmonds B, McKenzie F, Panoch JE, Wocial LD, Barnato AE, Frankel RM. "Doctor, what would you do?": physicians' responses to patient inquiries about periviable delivery. Patient Educ Couns. Jan 2015;98(1):49-54. doi:10.1016/j.pec.2014.09.014
17. Peters MD, Godfrey CM, Khalil H, McInerney P, Parker D, Soares CB. Guidance for conducting systematic scoping reviews. Int J Evid Based Healthc. Sep 2015;13(3):141-6. doi:10.1097/xeb.0000000000000050
18. Tricco AC, Lillie E, Zarin W, et al. PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. Ann Intern Med. Oct 2 2018;169(7):467-473. doi:10.7326/m18-0850
19. Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan-a web and mobile app for systematic reviews. Syst Rev. Dec 5 2016;5(1):210. doi:10.1186/s13643-016-0384-4

20. Roscigno CI, Savage TA, Kavanaugh K, et al. Divergent views of hope influencing communications between parents and hospital providers. Qual Health Res. Sep 2012;22(9):1232-46. doi:10.1177/1049732312449210
21. Kavanaugh K, Nantais-Smith LM, Savage T, Schim SM, Natarajan G. Extended family support for parents faced with life-support decisions for extremely premature infants. Neonatal Netw. Sep-Oct 2014;33(5):255-62. doi:10.1891/0730-0832.33.5.255
22. Kavanaugh K, Roscigno CI, Swanson KM, Savage TA, Kimura RE, Kilpatrick SJ. Perinatal palliative care: Parent perceptions of caring in interactions surrounding counseling for risk of delivering an extremely premature infant. Palliat Support Care. Apr 2015;13(2):145-55. doi:10.1017/s1478951513000874
23. Berman H, Koch PM, Freer JP, Craenen G. Comfort Care Request for Preterm Infant: Prescriptive Analysis. Am J Bioeth. Jan 2017;17(1):84-86. doi:10.1080/15265161.2016.1251635
24. Tucker Edmonds B, Savage TA, Kimura RE, et al. Prospective parents' perspectives on antenatal decision making for the anticipated birth of a periviable infant. J Matern Fetal Neonatal Med. Mar 2019;32(5):820-825. doi:10.1080/14767058.2017.1393066
25. French KB. Care of Extremely Small Premature Infants in the Neonatal Intensive Care Unit: A Parent's Perspective. Clin Perinatol. Jun 2017;44(2):275-282. doi:10.1016/j.clp.2017.01.008
26. 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. Mar 2019;23(3):422-430. doi:10.1007/s10995-018-02727-8
27. Jager S, Kavanaugh K, Hoffman S, Laitano T, Jeffries E, Tucker Edmonds B. Parents' Descriptions of Neonatal Palliation as a Treatment Option Prior to Periviable Delivery. J Perinat Neonatal Nurs. Apr/Jun 2020;34(2):178-185. doi:10.1097/jpn.0000000000000483
28. Eves MM, Danziger PD, Farrell RM, Cole CM. Conflicting Values: A Case Study in Patient Choice and Caregiver Perspectives. Narrat Inq Bioeth. Summer 2015;5(2):167-78. doi:10.1353/nib.2015.0054
29. Haward MF, Lorenz JM, Janvier A, Fischhoff B. Bereaved Parents: Insights for the Antenatal Consultation. Am J Perinatol. Jul 12 2021;doi:10.1055/s-0041-1731651
30. 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. Mar 2021;41(3):396-403. doi:10.1038/s41372-020-0739-5
31. 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. Jan-Mar 2011;25(1):52-60. doi:10.1097/JPN.0b013e31820377e5
32. Young E, Tsai E, O'Riordan A. A qualitative study of predelivery counselling for extreme prematurity. Paediatr Child Health. Oct 2012;17(8):432-6. doi:10.1093/pch/17.8.432
33. Pepper D, Rempel G, Austin W, Ceci C, Hendson L. More than information: a qualitative study of parents' perspectives on neonatal intensive care at the extremes of prematurity. Adv Neonatal Care. Oct 2012;12(5):303-9. doi:10.1097/ANC.0b013e318265b3d5
34. Hendriks MJ, Abraham A. End-of-Life Decision Making for Parents of Extremely Preterm Infants. J Obstet Gynecol Neonatal Nurs. Sep-Oct 2017;46(5):727-736. doi:10.1016/j.jogn.2017.06.006
35. Ursin L, Syltern J. Protect us from ourselves: Balancing the parental instinct of saving. Nurs Ethics. Aug 2020;27(5):1282-1296. doi:10.1177/0969733019871691
36. Kharrat A, Moore GP, Beckett S, Nicholls SG, Sampson M, Daboval T. Antenatal Consultations at Extreme Prematurity: A Systematic Review of Parent Communication Needs. J Pediatr. May 2018;196:109-115.e7. doi:10.1016/j.jpeds.2017.10.067
37. Shapiro N, Wachtel EV, Bailey SM, Espiritu MM. Implicit Physician Biases in Periviability Counseling. J Pediatr. Jun 2018;197:109-115.e1. doi:10.1016/j.jpeds.2018.01.070
38. Kunkel MD, Downs SM, Tucker Edmonds B. Influence of Maternal Factors in Neonatologists' Counseling for Periviable Pregnancies. Am J Perinatol. Jul 2017;34(8):787-794. doi:10.1055/s-0037-1598247
39. Feudtner C, Morrison W. The darkening veil of "do everything". Arch Pediatr Adolesc Med. Aug 2012;166(8):694-5. doi:10.1001/archpediatrics.2012.175

40. de Vries M, Fagerlin A, Witteman HO, Scherer LD. Combining deliberation and intuition in patient decision support. Patient Educ Couns. May 2013;91(2):154-60. doi:10.1016/j.pec.2012.11.016
41. Strick M, Papies EK. A Brief Mindfulness Exercise Promotes the Correspondence Between the Implicit Affiliation Motive and Goal Setting. Pers Soc Psychol Bull. May 2017;43(5):623-637. doi:10.1177/0146167217693611
42. Epstein RM, Gramling RE. What is shared in shared decision making? Complex decisions when the evidence is unclear. Med Care Res Rev. Feb 2013;70(1 Suppl):94s-112s. doi:10.1177/1077558712459216

Appendix s1: electronic search strategies (last updated 15th april 2022)

Pubmed search

("Fetal Viability"[Mesh] OR "Infant, Premature"[Mesh] OR "Infant, Very Low Birth Weight"[MeSH] OR "Premature Birth"[MesH] OR VLBW[tiab] OR ELBW[tiab] OR EPI[tiab] OR ((viability[tiab] OR viable[tiab] OR periviab*[tiab] OR prematur*[tiab] OR preterm*[tiab] OR Pre-Term*[tiab] OR Pre-Matur*[tiab]) AND (baby[tiab] OR babies[tiab] OR birth*[tiab] OR childbirth*[tiab] OR neonat*[tiab] OR infant*[tiab] OR newborn*[tiab] OR child*[tiab]))) AND

("Decision Making"[mh] OR "Counseling"[mh] OR Decision making[tiab] OR Counseling[tiab] OR Counselling[tiab] OR "Withholding Treatment"[mh] OR "Resuscitation"[mh] OR "Intensive Care, Neonatal"[mh] OR Withholding treatment*[tiab] OR Resuscitat*[tiab] OR Neonatal intensive care[tiab] OR Palliati*[tiab] OR Comfort care[tiab] OR Active care[tiab] OR Active treatment[tiab] OR NICU[tiab] OR Neonatal care[tiab]) AND

("Parents"[mh] OR "Pregnant Women"[mh] OR Parent*[tiab] OR Father*[tiab] OR Mother*[tiab] OR Pregnant woman[tiab] OR Pregnant women[tiab] OR Maternal[tiab] OR Paternal[tiab] OR Parental[tiab]) Filters: from 2010 - 2022

This search string was adapted to each database.

Table S2 characteristics of the included articles

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
1	Moro et al	2011	USA	To describe parents', nurses' and physician's perspectives on how parents make life support decisions for extremely premature infants prenatal through end-of-life period	Five mothers, four physicians, three registered nurses and one neonatal nurse practitioner All mothers gave birth between 23^0 and 24^5 weeks of GA of a liveborn infant who died later	Collective case study design using interviews and medical charts, single centre	Prenatal and postnatal 'little after birth' The interviews of end-of-life decisions were taken weeks to months after the death of the infant	Hope for survival Prevent pain and suffering Everything done Hope (general)
2	Young et al	2012	Canada	To gain insight on current practices from those most affected by the decisions made in the delivery room – the parents of the extreme premature babies	Ten mothers and six fathers of extremely premature infants (23-26-week of GA) who survived	Qualitative study using semi-structured interviews, single centre	All interviews <4 years after birth, except one	Potential complications of prematurity Suffering of their infant If they had known 'what was in store' would not have proceeded resuscitation
3	Roscigno et al	2012	USA	To report and evaluate parental and Healthcare providers descriptions of hope following counselling of parents at risk of delivering an extremely premature infant	40 mothers who were hospitalized for a threatened preterm delivery <26 week of GA, 14 of their partners and 71 healthcare providers (physicians, registered nurses and neonatal nurse practitioners)	Data was extracted from a (qualitative) longitudinal multiple case study using semi structured interview guides, multicentre	Prenatal and postnatal 'little after birth' Interviews about end-of-life decisions were taken weeks to months after death	The meaning of a child with a disability Hope for survival Religion (Gods hands), spirituality and faith Hope for a 'good outcome' Experiences of parents

Table S2 Continued

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
4	Pepper et al	2012	Canada	To describe and understand the decision-making process for parents whose infants were born and survived at the extremes of prematurity at 24 to 26 weeks' gestation	Seven parents representing five families	Qualitative, interpretive description, semi-structured interviews	Postnatal, not clearly identified how long after birth	Potential handicap Percentage for complications really high Let god decide, put faith in something else
5	Kavanaugh et al	2014	USA	To outline parental descriptions of extended family involvement and support surrounding decision making for their extremely preterm infant	54 parents; 40 mothers and 14 fathers of extremely premature infants 22-25^{6}-weeks of GA	Collective case study using data extracted from a large, longitudinal multiple collective case study design using semi-structured interviews	Prenatal and postnatal little after birth The interviews about end-of-life decisions were taken weeks to months after the death of the infant	Advice 'up to God' can be hurtful / not valuing life Rely on (relatives') Experiences of parents
6	Kavanaugh et al	2015	USA	To describe how parents at risk of delivering prematurely (<26 GA) interpreted the quality of their interpersonal interaction with Healthcare providers	54 parents; 40 mothers and 14 fathers of extremely premature infants 22-25^{6}-weeks of GA	Secondary analyses of data extracted from a large, longitudinal multiple collective case study design using semi-structured interviews	Prenatal and postnatal 'little after birth' The interviews about end-of-life decisions were taken weeks to months after the death of the infant	Doing what is right, what is natural hope Infants' best interest (versus family's interest)

Table S2 Continued

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
7	Eves et al	2015	USA	To highlight how bias may undermine the Healthcare providers ability to meet their obligation to enhance parent's autonomy and the moral distress they may experience when parental values do not align with their own	1 woman, 22^{3}-weeks pregnant, at risk for delivering prematurely	Case report	Prenatal	A degree of disability is inhumane Effect on family
8	Hendriks & Abraham	2017	Switzerland	To explore parental attitudes and values in the process of end-of-life decisions of extremely preterm infants (<28 weeks of GA)	Seven couples, five mothers and one father (n = 20) of extremely preterm infants (<28 weeks of GA) who were born alive and died in the delivery room or NICU	Qualitative study using in-depth, narrative interviews with semi-structured questions to clarify specific themes, single centre	Timeframe of 1-2 years after the infant's death	Protect against burden of NICU-treatment/prevent suffering Try everything
9	Berman et al	2017	USA	To analyse the different available ethical options to Healthcare providers and the impact those have on parents and their infant	One mother, 24 weeks pregnant, delivering prematurely	Case report	Prenatal	Caring for a disabled child Financial consequences
10	French	2017	USA	To describe different conversation the parents had with their healthcare providers about their daughter been born at 23^{+6} weeks of GA	One mother whose daughter was delivered at 23^{6} weeks of GA	Case report	Postnatal, not clearly identified how long after birth	Risk for long term morbidity Effect on marriage of hope for future children, medical debt To protect infants future

Table S2 Continued

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
11	Geurtzen et al	2019	Netherlands	To analyse parental preferences in prenatal counselling regarding the extremely premature infant	13 (pairs of) parents who experienced an extreme premature birth (both survivors and non-survivors) at 24 weeks of GA	Qualitative study using semi-structured interviews, multicentre	Interviews were taken 2-5 years after the counselling	Long term morbidity including QoL, participation in society, physical disabilities, mental disabilities, behavioural problems, No wrong or right, Risk-assessment for your infant and family Short term: length and intensity of (NICU) stay and complications Survival – mortality, To protect your baby, To take responsibility for your infant What you can handle as a parents
12	Edmonds et al	2019	USA	To examine prospective parental perceptions of management options and outcomes regarding threatened periviable delivery and the value's they apply in making a decision antenatal	54 prospective parents hospitalized for threatened periviable birth (22^0 to 25^6 weeks of GA)	Qualitative study, using data of prenatal interviews which were a part of a larger study, multicentre	Prenatal	Disability Healthy infant Protect against burden of NICU-treatment/ concert about pain and suffering Faith Responsibility for decision Doing what was best Giving a fighting chance Experiences

Table S2 Continued

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
13	Lynch et al	2019	USA	To help Healthcare providers determine how to support their patients in medically complex and emotionally laden decisions by understanding parental experiences of their periviable deliveries	Ten women who delivered between 22^0-25^0 weeks of GA	Qualitative study using open-ended, semi-structured interviews, single centre	24h-12 days after delivery	Survival Families interest versus infants interest Try everything What is best for her
14	Ursin & Syltern	2019	Norway	Discussing the role of parents in neonatal decision-making, based on the following research question: Should parents decide whether to provide lifesaving treatment when their child is born at the limit of viability?	12 parents of children born at 23–24 weeks of GA, one at 34 weeks of GA (threatening birth at 22 weeks of GA)	Qualitative study using structured interview format, multicentre	Postnatal, not clearly identified how long after birth	Not wanting this life for the infant Survival Long term outcomes for the family system Everything done Responsible for decision/instinct of saving/protecting your child Wanting the best for the child
15	Jager et al	2020	USA	To explore the language pregnant woman and important others use when discussing comfort care as an option to treat their extremely premature infant	30 pregnant woman (between 22^0 and 24^6 weeks of GA) and their 16 important others	Secondary analysis of data collected as a part of prospective qualitative and quantitative study, multicentre	Prenatal, interviews were taken after receiving antenatal consultation from the NICU-team	Not wanting the child to have a low quality of life Chance of survival Protect against burden of NICU-treatment; not be in pain and suffering Having faith Not giving up hope Giving the baby a chance

Table S2 Continued

#	Authors	Year	Country	Aim of study	Study population and sample size	Methodology	Timing of researching the parents	*Extracted key findings (parental perspectives) concerning our study objectives*
16	Haward et al	2021	USA	To explore parental experiences of extremely preterm infant loss in the delivery room an perspectives about prenatal consultation	13 participants who experienced loss of their infant	Qualitative study using semi-structured interviews, multicentre	Postnatal, not clearly identified how long after birth	Long term outcomes for the child/(future) quality of life Survival Protect against pain and suffering Long-term outcomes for the family system Faith Hope versus realism Doing what is right Experiences
17	Tucker Edmonds et al	2021	USA	To explore perceptions of pain/ suffering, disability and coping by race among pregnant women facing the threat of a periviable delivery	30 pregnant women facing the threat of a periviable delivery (22^0-24^6 weeks of GA)	Qualitative study using semi-structured interviews, multicentre	Prenatal	Long term outcomes for the child/concerns for (long term) quality of life Survival Protect against burden of NICU-treatment Faith Everything done Giving a (fighting) chance/strength to fight

Abbreviations: GA: Gestational Age, NICU: Neonatal Intensive Care Unit, ELBW: Extremely Low Birth Weight