



UvA-DARE (Digital Academic Repository)

How matters

End-of-life communication with family members of critically ill neonates, children, and adults

Prins, S.

Publication date

2024

[Link to publication](#)

Citation for published version (APA):

Prins, S. (2024). *How matters: End-of-life communication with family members of critically ill neonates, children, and adults*. [Thesis, fully internal, Universiteit van Amsterdam].

General rights

It is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), other than for strictly personal, individual use, unless the work is under an open content license (like Creative Commons).

Disclaimer/Complaints regulations

If you believe that digital publication of certain material infringes any of your rights or (privacy) interests, please let the Library know, stating your reasons. In case of a legitimate complaint, the Library will make the material inaccessible and/or remove it from the website. Please Ask the Library: <https://uba.uva.nl/en/contact>, or a letter to: Library of the University of Amsterdam, Secretariat, Singel 425, 1012 WP Amsterdam, The Netherlands. You will be contacted as soon as possible.



CHAPTER 2

HOW PHYSICIANS DISCUSS UNCERTAINTY WITH PARENTS OF CRITICALLY ILL NEONATES AND CHILDREN IN INTENSIVE CARE UNITS

This chapter is published as:

Prins S, Linn AJ, van Kaam AHL, van de Loo M, van Woensel JBM, van Heerde M, Dijk PH, Kneyber MCJ, de Hoog M, Simons SHP, Akkermans A, Smets EMA, Hillen MA, de Vos MA (2022) How physicians discuss uncertainty with parents in intensive care units. *Pediatrics* 149(6):e2021055980.
<https://doi.org/10.1542/peds.2021-055980>

ABSTRACT

What's known on the subject

Prevailing uncertainties complicate decision-making in the neonatal and pediatric intensive care unit. They also appear to negatively affect patient- and parent-related health outcomes. Adequately discussing uncertainty with parents is pivotal yet challenging for physicians.

What this study adds

This explorative study shows that physicians use a wide array of strategies to discuss uncertainty with parents. These strategies vary throughout the child's illness and treatment trajectory. Our insights will help physicians to better tailor their approach to parental needs.

Background and objectives

Physicians and parents of critically ill neonates and children receiving intensive care have to make decisions on the child's behalf. Throughout the child's illness and treatment trajectory, adequately discussing uncertainties with parents is pivotal, as this enhances the quality of the decision-making process and may positively affect the child's and parents' well-being. We investigated how physicians discuss uncertainty with parents and how this discussion evolves over time during the trajectory.

Methods

We asked physicians working in the neonatal intensive care unit (NICU) and pediatric intensive care unit (PICU) of three university medical centers to audio record their conversations with parents of critically ill children from the moment doubts arose whether treatment was in the child's best interests. We qualitatively coded and analyzed the anonymized transcripts, thereby using the software tool MAXQDA 2020.

Results

Physicians were found to adapt the way they discussed uncertainty with parents to the specific phase of the child's illness and treatment trajectory. When treatment options were still available, physicians primarily focused on uncertainty related to diagnostic procedures, treatment options and associated risks and effects. Particularly when the child's death was imminent, physicians had less 'scientific' guidance to offer. They

eliminated most uncertainty and primarily addressed practical uncertainties regarding the child's dying process to offer parents guidance.

Conclusions

Our insights may increase physicians' awareness and enhance their skills in discussing uncertainties with parents tailored to the phase of the child's illness and treatment trajectory and to parental needs in each specific phase.

INTRODUCTION

In the neonatal intensive care unit (NICU) and pediatric intensive care unit (PICU), physicians and parents often have to engage in complex decision-making on the child's behalf.^{1,2} Parents need to be well-informed about their child's illness, prognosis, and available treatment options to enable participation in the decision-making process. Thus, physicians are tasked to inform parents fully, clearly, and honestly, which includes discussing any potential or existing uncertainties.³⁻⁵ Such uncertainties may include risks versus benefits of diagnostic or therapeutic procedures, the child's well-being in the short and longer term, and dilemmas about the utility of life-sustaining treatment (LST) versus end of life.^{1,3,6-9} Such conversations can be difficult as uncertainties may diminish parents' sense of hope and increase their emotional distress. Uncertainty may also provoke avoidance of decision-making.^{8,10-16} Physicians previously expressed reluctance to communicate uncertainties, due to concerns about its negative effects for parents and themselves.^{8,13,14,17-20} However, growing evidence suggests that adequately discussing uncertainties with parents enhances decision-making and well-being of critically ill children and their parents, for instance by giving parents a sense of empowerment.^{3,11,21-28}

More insights are needed into *how* physicians discuss uncertainties with parents and how such discussions evolve over time, especially in cases in which the child's health further deteriorates in the NICU and PICU.^{10,29-31} We aim to prospectively investigate (1) to which topics physicians' discussion of uncertainty pertains in physician-parent conversations in the NICU and PICU, (2) which communicative strategies physicians use to discuss uncertainty with parents, and (3) how the discussion of uncertainty develops over time.

METHOD

Procedure and participants

This explorative qualitative study was part of a larger research project about communication and decision-making in family conferences (henceforth: conversations) (FamICom).³² Audio-recordings were made of conversations between physicians (neonatologists, pediatric intensivists and/or consulting specialists), nurses and parents of children admitted to the NICU and PICU of three university medical centers in the Netherlands. Physicians and nurses from the six participating intensive care units received oral and written study information and signed informed consent before participation. All physicians and all nurses, except for one NICU nurse, consented.

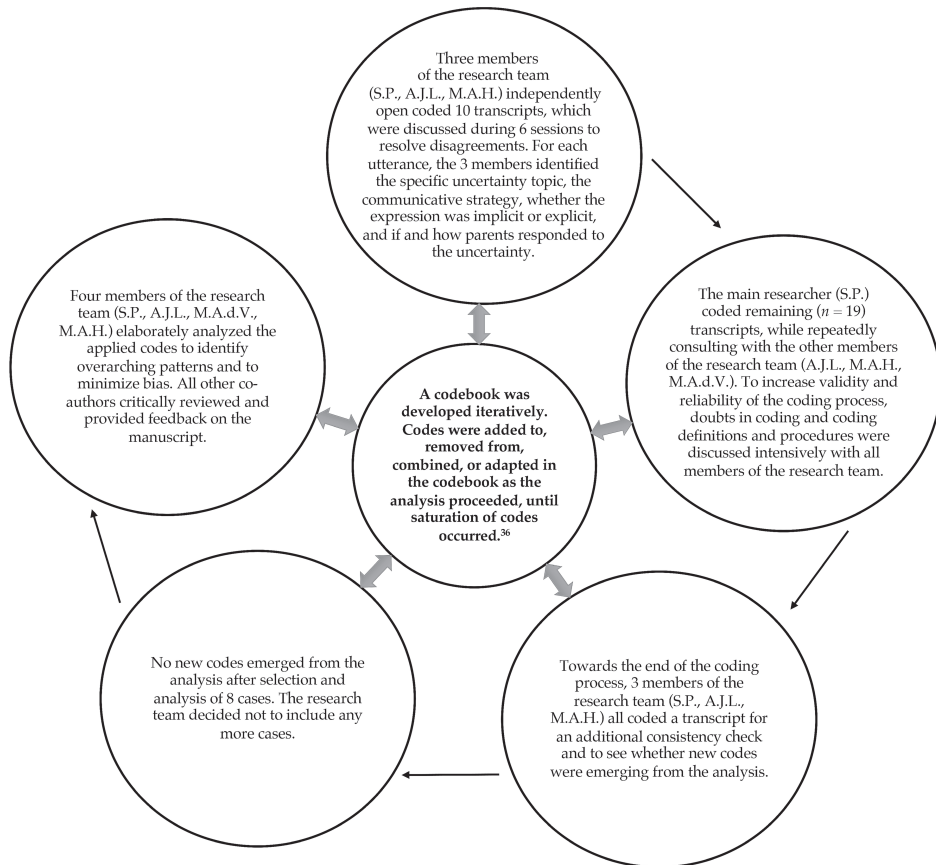
Parents were eligible as soon as they themselves or the medical team expressed doubts whether continuing LST was in the child's best interests. Parents were personally approached in the clinic and informed about the study by the attending physician or a member of the research team. If willing to participate, parents provided verbal and written consent. All formal conversations between physicians and parents were audio-recorded until a final decision was made to either continue or to discontinue LST. The University's Institutional Review Board approved the study protocol on behalf of all centers (W17_475 # 17.548).

Sample selection

The full dataset consisted of the audio-recordings of conversations regarding 36 children (NICU $n = 19$; PICU $n = 17$). Maximum variation was sought regarding the patient's age, sex, diagnosis, disease progression and course of treatment, and the parents' ethnic background, level of education, and religious beliefs. In this study, we only included cases in which at least three conversations were audio-recorded. This enabled us to explore how the discussion of uncertainty evolved over time.

Qualitative analysis

Audio-recordings were transcribed verbatim and anonymized. The transcripts were then analyzed using the software tool MAXQDA 2020.³³ All transcripts were carefully read and for each utterance of physicians that contained any aspect of uncertainty, three coders (S.P., A.J.L., M.A.H) identified the *topic*, the *communicative strategy*, whether the expression was *implicit* or *explicit*, and if and how *parents responded* to the uncertainty, until data saturation was reached.³⁴⁻³⁶ In accordance with the widely accepted definition by Han, we defined uncertainty as 'the subjective consciousness of ignorance', which implies that uncertainty concerns a metacognitive personal awareness of one's lack of knowledge.¹⁰ Details of our steps of coding and explorative analysis are provided in Figure 1.

Figure 1. Our steps of coding and analysis of uncertainties

RESULTS

In total, 29 conversations were analyzed regarding 8 patients (NICU $n = 4$; PICU $n = 4$, Table 1). Sixteen parents and 29 physicians participated in these conversations. Although nurses were present in most conversations, they did not actively participate in the discussion of uncertainties.

Physicians rarely addressed uncertainty explicitly, for instance by stating that they 'did not know' something. These few explicit expressions mainly concerned practical topics like the moment when test results would be known. Yet, physicians predominantly discussed uncertainty in implicit ways, for instance by using terms as 'it seems' or 'it could be'.

Table 1. Patient case characteristics^a

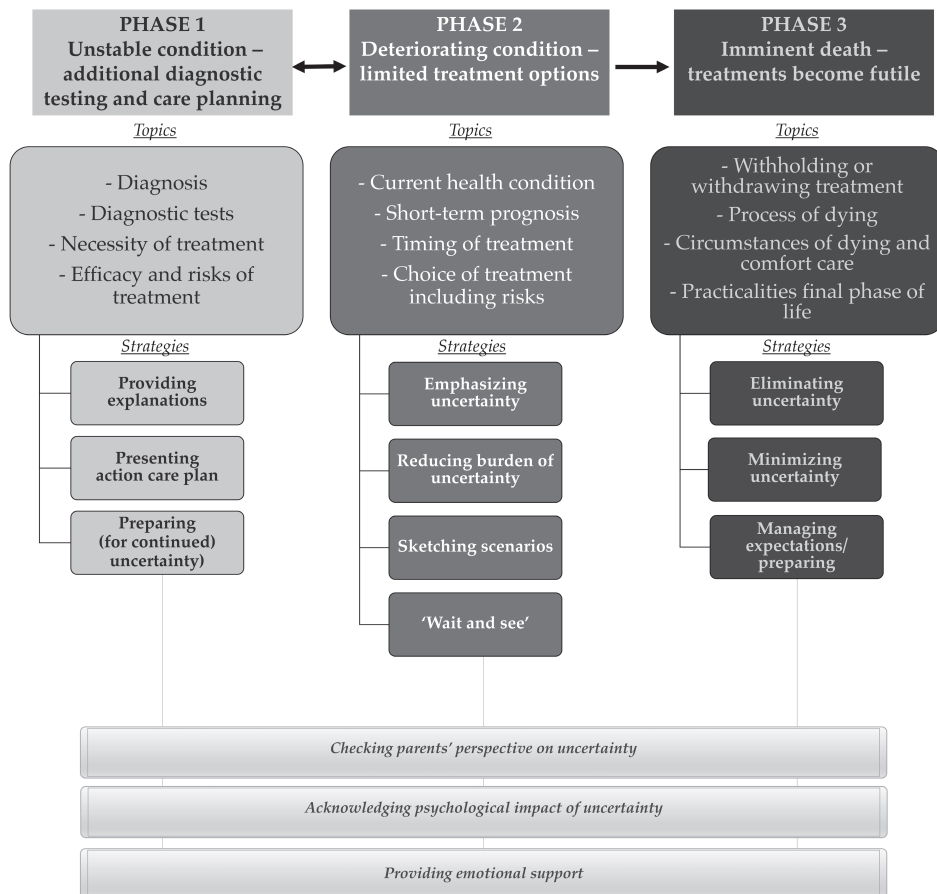
# Case	Description
#N1	A prematurely born boy admitted to the neonatal intensive care unit, who seemed stable, but suddenly deteriorated due to unexplainable kidney failure and would die soon.
Data:	Three conversations between both parents with a migration background, two neonatologists and two nurses.
#N2	A prematurely born girl admitted to the neonatal intensive care unit, who was transferred to medium care at another hospital due to sudden improvement.
Data:	Three conversations between a Dutch father and immigrant mother and two neonatologists.
#N3	A prematurely born boy admitted to the neonatal intensive care unit. The child's twin brother was also admitted to the neonatal intensive care unit. Whereas the brother was doing well, this child's situation remained unstable and deteriorated.
Data:	Three conversations between Dutch parents, and three neonatologists.
#N4	A prematurely born girl admitted to the neonatal intensive care unit. The child's situation remained unchanged, i.e., did not improve but also did not get worse, while receiving life-sustaining treatment.
Data:	Four conversations between Dutch parents and two neonatologists.
#P1	A male toddler (age range 1-4 years) with a congenital disorder, admitted to the pediatric intensive care unit, who deteriorated and died.
Data:	Six conversations between immigrant parents and three pediatric intensivists, two pediatricians, two pediatric neurologists, a metabolic pediatrician, seven nurses and a social worker.
#P2	A teenage boy (age range 12-16 years) with a congenital disorder, admitted to the pediatric intensive care unit. During earlier hospital admissions, the boy had a good quality of life, but now deteriorated and died.
Data:	Three conversations between Dutch parents and the patient's brother, and a pediatrician, two pediatric intensivists, an anesthesiologist, a nurse, a medical educationalist, and a social worker.
#P3	A girl (age range 16-21 years) with a congenital disorder, admitted to the pediatric intensive care unit. Parents quickly decided that the child suffered too much, and they requested palliative care, to which physicians eventually agreed. Parents took their child home for the final phase of life.
Data:	Four conversations between Dutch parents, and an anesthetist-pediatric intensivist, two pediatric intensivists, a pediatric neurologist, two pediatricians, and two nurses.
#P4	A girl (age range 4-12 years) with a congenital disorder, admitted to the pediatric intensive care unit. Physicians were committed to do everything possible, although the child's situation seemed unexplainable. Child had been admitted to the intensive care unit for already five months. Situation slightly improved and the plan was to transfer the child to a special care unit, but this seemed too risky.
Data:	Four conversations between Dutch parents and two pediatricians.

^a Due to privacy regulations, we report age ranges instead of exact ages.

The uncertainties that physicians discussed concerned a wide array of topics, ranging from the child's diagnosis and (short-term) prognosis to the content, timing and possible risks of diagnostic tests and treatment options, and - eventually - the process of dying.

We distinguished three distinct phases in children’s illness and treatment trajectory, each associated with specific topics of uncertainty and identifiable communicative strategies which physicians used to discuss these topics (Figure 2). Although the term ‘strategy’ might imply intentional use, we were not able to assess how intuitive or intentional physicians’ use of strategies was. In most, but not all, cases the three phases occurred sequentially. In some conversations, we identified three additional strategies that appeared to be independent from the three identified phases.

Figure 2. Main topics to which uncertainty pertained and main strategies to discuss uncertainty with parents per phase of children’s illness and treatment trajectory



Phase 1: Unstable condition – Additional diagnostic testing and care planning

In this first phase, the condition of the child was typically unstable, while underlying causes for such instability were unknown in most cases. Physicians’ communication

mainly centered around short-term uncertainties, for example how the child's condition would develop in the coming days.

Physicians addressed these uncertainties using communicative strategies such as providing a range of **possible explanations** for the causes of the child's current situation, or by suggesting a **most likely explanation**.

#N1.1, Physician: "His problems may be explained by a coarctation, and that when muscle tissue around the duct contracts, it also partly narrows his aorta."

Physicians often accompanied these explanations by **presenting a short-term action plan**. This plan included further diagnostics or treatments or a proposition to consult specialized colleagues.

#N1.2, Physician: "We want to do more tests to find out what causes his acute kidney failure. We want to do another scan, give extra fluids and if that doesn't work, we will give diuretics."

In presenting such plans, physicians also **prepared parents for subsequent uncertainties**, for example about the feasibility and risks of diagnostic tests, as illustrated in the following citation:

#P1.1, Physician: "Doing an MRI is challenging, because it would be difficult to manage his breathing and oxygen levels. Currently, he is unable to breathe without the mask. Thus, we would have to intubate and sedate him for only an MRI."

Parents hardly responded when physicians addressed such kinds of uncertainties in this phase. In the few instances in which they did respond, parents requested additional information, or asked for clarification. Notably, in one PICU case, parents took the lead from the first conversation on, and clearly indicated their preferences regarding withdrawing their child's treatment. Only in this case, the physician addressed the process and circumstances of dying in an early phase, including important practical uncertainties (e.g., housing situation and place of last phase of life).

Phase 2: Deteriorating condition –Limited treatment options

In phase two, the child's situation typically deteriorated, where reasons for deterioration often remained unclear and treatment options became limited. The primary treatment goal shifted toward protecting the child from suffering. During these conversations, physicians stated that "something" needed to be done, but that it also became more and more uncertain *what* could be done exactly. Thereby, a balance of whether the remaining options would not cause more harm than benefit in the short or long-term were discussed.

In this phase, physicians used more diverse strategies to address uncertainty than in phase one and three.

Primarily, physicians **emphasized uncertainties**, thereby making parents aware of the possible negative outcomes of tests and treatments.

#N3.3, Physician: “But I want you to realize that still anything can happen. Because he will be undergoing heart surgery which comes with a lot of risks.”

Additionally, physicians stressed their increased worries about the child’s condition. In this way, they put even more emphasis on how problematic and uncertain the situation had become.

Simultaneously, physicians appeared to **reduce the burden of the uncertainty** for parents, for instance by reassuring them about the quality of care their child was receiving. In the following example, the physician had just explained to the parents that an intubation is a risky procedure that could also alleviate the child’s suffering:

#P2.2, Physician: “The procedure of intubating is difficult and carries the risk that he will die during this procedure. But I assure you that you are in the right place. We have all kinds of physicians here who are highly experienced in intubating children in complex circumstances.”

Particularly in the NICU setting, physicians addressed uncertainties by **sketching different possible scenarios**. This was done in three different ways. First, physicians sometimes only sketched a positive scenario, seemingly in an effort to help parents retain their hope. Second, they sketched a negative or worst-case scenario. Physicians sometimes did so in reaction to parents’ utterances of hope. Here, it appeared a way of counterbalancing parents’ unrealistic hopes. Third, physicians neutrally listed multiple possible scenarios or outlined the most likely scenario that might happen.

Interestingly, when sketching a negative scenario, physicians were more likely to generalize uncertainty instead of referring to the situation of this specific child.

#N4.3, Physician: “It appears to go well right after the breathing tube has been taken out; but we often see that children slowly deteriorate later on.”

Finally, physicians addressed uncertainty in this second phase by stating that they would **‘wait and see’** how the child’s situation would develop over the coming hours or days. This is in stark contrast to the strategy used in phase one, where physicians would propose a concrete action plan to diminish uncertainty.

Again, very few parents directly responded to physicians' various strategies to address uncertainty. If parents did respond, they expressed their understanding or stressed their hope that things would still work out for their child.

Phase 3: Imminent death – Treatments become futile

In this last phase, appropriate therapeutic treatment options had been exhausted, and it became clear that the child would die soon. Uncertainties in this phase especially entailed decisions whether to continue or withdraw LST, the implementation and timing of such decisions, and other practical uncertainties, such as how long the process of dying lasts and how parents could best support their child during this process.

Physicians' main strategy in this phase was to **eliminate any remaining uncertainty** about whether treatment had truly become futile and about the inevitability of the child's death.

P2.3, Physician: *"We don't think it's fair to do anything to him anymore [...]. Uhm, it would not be appropriate to continue mechanical ventilation. Because we know for sure that he will die."*

Moreover, physicians **minimized uncertainty** by reassuring parents that they would do everything they could to prevent the child from suffering.

P3.2, Physician: *"The most important aspect is that we are going to make sure she is comfortable so that she will not suffer."*

Finally, physicians thoroughly **prepared** parents for the dying process of their child, thereby **managing parents' expectations**, as illustrated in the following example:

P1.6, Physician: *"Things such as the ventilator, we will stop them. We do not know for sure how he will respond to that; whether he will keep breathing – I expect that he won't."*

In response to the addressed uncertainties, a few parents requested more practical information, while others reacted emotionally, for example by expressing their feelings of fear or guilt or by starting to cry.

Strategies throughout the whole trajectory

We observed three strategies that physicians used to address uncertainty, which were not phase-specific but occurred in every phase of the illness and treatment trajectory. Although these strategies occurred quite rarely, they appeared to help parents express their needs, as underlined by more explicit responses from parents.

The first strategy consisted of physicians **checking parents' perspective on the uncertainty**.

#P4.1, Physician: "*We need to take time to see how the situation will develop. How do you feel about that?*"

The second strategy consisted of **acknowledging the psychological impact of uncertainty**, by explicitly mentioning how difficult the uncertainty had to be for parents. The last strategy consisted of **providing emotional support** to help parents cope with the high amount of uncertainty, for instance by stating:

#P1.5, Physician: "*We are here for you if you need us.*"

DISCUSSION

In this study we explored how physicians discuss uncertainty with parents of critically ill children admitted to the NICU and PICU, and how this discussion evolves over time, by inductively coding and analyzing successive audio-recorded conversations.

We identified three distinct phases with a clear relationship between the uncertainty topics physicians addressed, the communicative strategies they used to discuss these topics, and how these combinations of topics and strategies evolved during the child's illness and treatment trajectory. These three phases align with the 'Phases of Illness' previously classified in palliative care.^{37,38} Our results contribute to the literature by showing that physicians adapt the way they discuss uncertainty with parents to the specific phase of the child's illness and treatment trajectory, either consciously or unconsciously.

In previous studies, physicians were found to focus almost exclusively on 'scientific uncertainty', i.e., uncertainty related to diagnosis, prognosis, treatment options and the possible risks and effects of these options.^{3,30,39-43} Physicians in our study appeared to discuss a wider array of uncertainty topics, including practical and personal uncertainties. Particularly in the last phase, physicians had less 'scientific' guidance to offer, and primarily addressed practical uncertainties regarding the child's dying process to still offer parents some guidance in coping with these uncertainties.

In the literature, three overarching strategies are presented in how physicians (may) address uncertainty with patients or their representatives: (1) preparing for the discussion of uncertainty, e.g., explicitly warning for uncertain outcomes *before* initiating diagnostic or therapeutic procedures, (2) informing about uncertainty, and (3) helping to deal with uncertainty, e.g., by providing emotional support.^{14,44,45} The strategies identified in the

current study mainly fall under the overarching strategy 2, rarely under strategy 3, and never under strategy 1. However, recent studies indicate that the needs and well-being of caregivers are best met by combining information-oriented strategies with coping-oriented strategies.^{9,18,22,46-50} Studies which retrospectively investigated parents' experiences during their child's stay in the NICU or PICU, concluded that openly discussing uncertainty positively affected parents' well-being and their (preparation for) bereavement, but only if it was combined with sufficient emotional support.^{4,9,31,50-53} By using information-oriented strategies, physicians may seem to solely focus on cognitive aspects, and neglect emotional aspects.⁹ Our study shows that this is not always the case. By using information-oriented strategies like sketching scenarios, physicians seemed to not only inform parents about the current uncertainties, but also to make these uncertainties bearable for parents and help them to prepare for potential worst-case scenarios. This may well improve their emotional coping on the longer term.⁵⁴

In none of the conversations, physicians explicitly asked parents about their information and emotional needs. Parents, for their part, hardly gave any explicit reaction to the uncertainties presented to them. We hypothesize that such limited or even lack of dialogue made it harder for physicians to tailor their communication strategies to the specific needs and wishes of individual parents. These needs and wishes may as well change over the course of their child's illness and treatment trajectory.³⁸ The three additional strategies we rarely found (i.e., checking parents' perspective on the uncertainty, acknowledging the psychological impact of uncertainty and providing emotional support), may be promising ways to help parents express their needs besides receiving sufficient guidance and support.

We found that physicians rarely disclosed prognostic uncertainty, in contrast to other topics of uncertainty. It may well be that physicians hesitated to explicitly share prognostic uncertainty out of fear to increase parents' anxiety and diminish their hope and trust.^{17,55,56} In neonatal and pediatric intensive care, prognostication may be particularly challenging because of the high unpredictability of how an individual child will react to treatments and to what extent he or she will recover.^{57,58} Yet, failing to timely and clearly address uncertain prognosis can lead to false hope or misunderstandings among parents.^{18,23} Rather than by nondisclosure, physicians could provide prognostic information in line with parents' information needs at that point in time, thereby also honestly explaining the limits of their prognosis.^{18,38,44,56,59,60}

Recommendations presented in figure 3 are a preliminary effort to support physicians in discussing uncertainty with parents.

Figure 3. Practical recommendations for discussing uncertainty

- 1 A **wide range of strategies** is available for physicians to discuss uncertainty with parents of critically ill children in and over the phases of the child's illness and treatment trajectory.
- 2 Physicians may use their **awareness of the three identified phases** to choose how to discuss uncertainty with parents throughout the child's illness and treatment trajectory.
- 3 Physicians could **explicitly explore** parents' individual responses to uncertainty, enabling them to tailor their discussion of uncertainty to parents' needs.
- 4 Using affective strategies when discussing uncertainty – i.e., **checking and acknowledging the impact of uncertainty** and **providing emotional support** – might facilitate parents in expressing their needs and help them in dealing with the conveyed uncertainty.
- 5 Particularly when appropriate treatment options are still available, physicians could consider discussing uncertainty in a **more explicit** way as this may help them to manage parents' expectations and facilitate prognostic awareness.
- 6 When death is imminent, **addressing practical uncertainties** – e.g., by explaining the practicalities of the last moments of the child's life in detail – could offer parents helpful guidance and support.
- 7 In the child's last phase of life, **eliminating any remaining uncertainty** may help parents accept that nothing more can be done, and that death is inevitable.

Although presenting novel and in-depth insights, our study has several limitations. First, our study took place in the Netherlands, where end-of-life practice allows withdrawal of life support. More research is needed to investigate whether and how discussions of uncertainty might be different in other countries with different regulations and practices. Second, in this study we included only cases involving three or more conversations to explore the development of uncertainty. Consequently, in the PICU, no parents were included whose otherwise healthy child had suddenly fallen critically ill. In future research, it would be of interest to investigate how discussions of uncertainty in such acute situations might be different. Third, in the context of our study, parents encountered a relatively large number of different physicians throughout the care trajectory. Such lack of continuity has been suggested to negatively affect the patient-provider relationship and information transfer.⁶¹⁻⁶⁴ Further research is needed to investigate whether the discussion of uncertainty develops differently in settings with more provider continuity. Fourth, we were unable to test effects or draw conclusions about how parents experienced physicians' communicative strategies about uncertainties. Previous studies have indicated that how uncertainty is communicated may positively or negatively affect patients' and families' levels of satisfaction and their

trust in the competence of healthcare providers.^{41,42,65-68} Last, physicians' intentions when addressing uncertainty could only be implicitly inferred from the context. We do not know whether physicians strategically made use of specific strategies or did this rather intuitively. Previous studies found that physicians sometimes deliberately use uncertainty to steer parents towards a certain decision.⁶⁹ Future research should address these limitations. We thereby suggest a mixed methods design in which audio- or video-recordings of actual conversations are combined with evaluative interviews with parents and physicians. In this way, a better insight can be obtained whether physicians use strategies deliberately and whether parents experience these strategies as helpful in the short and longer term.

Conclusions

This study thoroughly explored how physicians address uncertainties in complex and emotionally charged conversations in neonatal and pediatric intensive care. Physicians used a wide array of strategies to discuss uncertainty with parents which they adapt to the specific phase of the child's illness and treatment trajectory. Physicians rarely checked parents' needs. They did not explore which strategy parents found helpful at that point in time. Our insights may help physicians in becoming more aware of how they actually discuss uncertainties with parents and how they can improve this important part of their communication.

Acknowledgments

We are extremely grateful for the trust of the families, physicians and nurses who participated in this study. Furthermore, we would like to thank Sandra Dijkstra, Annelies Olthuis, Joke Dunk, Odile Frauenfelder, Maartje Harmelink and Joyce Lamerichs for their invaluable assistance in the data collection phase of this study. We thank Vicky Lehmann for correcting the English manuscript.

Author contributions

Ms Prins conceptualized and designed the study, collected, analyzed, and interpreted the data, drafted and finalized the initial manuscript and figures, and reviewed and revised the manuscript; Drs Linn and Hillen conceptualized and designed the study, analyzed and interpreted the data, drafted the initial manuscript, and critically reviewed and revised the manuscript; Ms Akkermans collected data, conceptualized and designed the study, and reviewed and revised the manuscript; Drs van Kaam, van de Loo, van Woensel, van Heerde, Dijk, Kneyber, de Hoog, Simons, and Smets conceptualized and designed the study and critically reviewed and revised the manuscript for important intellectual content; Dr de Vos collected, coordinated, and supervised data collection, conceptualized and designed the study, analyzed and interpreted the data, drafted the initial manuscript, and critically reviewed and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Declaration of conflicting interests

The authors have indicated they have no conflicts of interest relevant to this article to disclose.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This study is part of the research project 'FamICom', which was supported by ZonMw [project number 844001316]. ZonMw is the Dutch organization for healthcare research and innovation.

Data management and sharing

Data are available upon request.

REFERENCES

1. Bennett RA, LeBaron VT. Parental Perspectives on Roles in End-of-Life Decision Making in the Pediatric Intensive Care Unit: An Integrative Review. *Journal of Pediatric Nursing*. 2019;46:18-25. doi:10.1016/j.pedn.2019.02.029
2. Davidson JE, Aslakson RA, Long AC, et al. Guidelines for Family-Centered Care in the Neonatal, Pediatric, and Adult ICU. *Critical Care Medicine*. 2017;45(1):103-128. doi:10.1097/CCM.0000000000002169
3. Han PKJ, Klein WMP, Arora NK. Varieties of uncertainty in health care: a conceptual taxonomy. *Med Decis Making*. 2011;31(6):828-838. doi:10.1177/0272989X11393976
4. October TW, Fisher KR, Feudtner C, Hinds PS. The Parent Perspective: "Being a Good Parent" When Making Critical Decisions in the PICU. *Pediatric Critical Care Medicine*. 2014;15(4):291-298. doi:10.1097/PCC.0000000000000076
5. Brooten D, Youngblut JM, Caicedo C, Dankanich J. Parents: Wish I had done, wish I had not done, and coping after child NICU/PICU death. *Journal of the American Association of Nurse Practitioners*. 2019;31(3):175-183. doi:10.1097/JXX.0000000000000110
6. Hickman RL, Pinto MD. Advance directives lessen the decisional burden of surrogate decision-making for the chronically critically ill. *Journal of Clinical Nursing*. 2014;23(5-6):756-765. doi:https://doi.org/10.1111/jocn.12427
7. Cuttini M, Nadai M, Kaminski M, et al. End-of-life decisions in neonatal intensive care: Physicians' self-reported practices in seven European countries. *The Lancet*. 2000;355:2112-2118. doi:10.1016/S0140-6736(00)02378-3
8. Eden LM, Callister LC. Parent Involvement in End-of-Life Care and Decision Making in the Newborn Intensive Care Unit: An Integrative Review. *J Perinat Educ*. 2010;19(1):29-39. doi:10.1624/105812410X481546
9. Drach LL, Hansen DA, King TM, Sibinga EMS. Communication between neonatologists and parents when prognosis is uncertain. *J Perinatol*. 2020;40(9):1412-1422. doi:10.1038/s41372-020-0673-6
10. Han PKJ. Conceptual, Methodological, and Ethical Problems in Communicating Uncertainty in Clinical Evidence. *Med Care Res Rev*. 2013;70(1_suppl):14S-36S. doi:10.1177/1077558712459361
11. Hillen MA, Gutheil CM, Strout TD, Smets EMA, Han PKJ. Tolerance of uncertainty: Conceptual analysis, integrative model, and implications for healthcare. *Social Science & Medicine*. 2017;180:62-75. doi:10.1016/j.socscimed.2017.03.024
12. Kochen EM, Teunissen SCCM, Boelen PA, et al. Challenges in preloss care to parents facing their child's end-of-life: A qualitative study from the clinicians perspective. *Academic Pediatrics*. Published online August 27, 2021. doi:10.1016/j.acap.2021.08.015
13. Parascandola M, Hawkins JS, Danis M. Patient Autonomy and the Challenge of Clinical Uncertainty. *Kennedy Institute of Ethics Journal*. 2002;12(3):245-264. doi:10.1353/ken.2002.0018
14. Simpkin AL, Armstrong KA. Communicating Uncertainty: A Narrative Review and Framework for Future Research. *J GEN INTERN MED*. 2019;34(11):2586-2591. doi:10.1007/s11606-019-04860-8
15. October TW, Jones AH, Greenlick Michals H, Hebert LM, Jiang J, Wang J. Parental Conflict, Regret, and Short-term Impact on Quality of Life in Tracheostomy Decision-Making. *Pediatric Critical Care Medicine*. 2020;21(2):136-142. doi:10.1097/PCC.0000000000002109
16. Alaradi M. Predictors of uncertainty, stress, anxiety, and depressive symptoms of parents of preterm infants in the neonatal intensive care unit. *Electronic Theses and Dissertations*. Published online August 1, 2014. doi:10.18297/etd/25

17. Apatira L, Boyd EA, Malvar G, et al. Hope, Truth, and Preparing for Death: Perspectives of Surrogate Decision Makers. *Ann Intern Med*. 2008;149(12):861-868. doi:10.7326/0003-4819-149-12-200812160-00005
18. Brouwer MA, Maeckelberghe ELM, Heide A van der, Hein IM, Verhagen EAEE. Breaking bad news: What parents would like you to know. *Archives of Disease in Childhood*. 2021;106(3):276-281. doi:10.1136/archdischild-2019-318398
19. Politi MC, Légaré F. Physicians' reactions to uncertainty in the context of shared decision making. *Patient Educ Couns*. 2010;80(2):155-157. doi:10.1016/j.pec.2009.10.030
20. Katz J. Why Doctors Don't Disclose Uncertainty. *The Hastings Center Report*. 1984;14(1):35-44. doi:10.2307/3560848
21. Haward MF, Murphy RO, Lorenz JM. Message Framing and Perinatal Decisions. *Pediatrics*. 2008;122(1):109-118. doi:10.1542/peds.2007-0620
22. Labrie NHM, van Veenendaal NR, Ludolph RA, Ket JCF, van der Schoor SRD, van Kempen AAMW. Effects of parent-provider communication during infant hospitalization in the NICU on parents: A systematic review with meta-synthesis and narrative synthesis. *Patient Educ Couns*. 2021;104(7):1526-1552. doi:10.1016/j.pec.2021.04.023
23. Meert KL, Eggly S, Pollack M, et al. Parents' perspectives on physician-parent communication near the time of a child's death in the pediatric intensive care unit. *Pediatr Crit Care Med*. 2008;9(1):2-7. doi:10.1097/01.PCC.0000298644.13882.88
24. Schaefer KG, Block SD. Physician communication with families in the ICU: Evidence-based strategies for improvement. *Current Opinion in Critical Care*. 2009;15(6):569-577. doi:10.1097/MCC.0b013e328332f524
25. Wong P, Liamputtong P, Koch S, Rawson H. Barriers to regaining control within a constructivist grounded theory of family resilience in ICU: Living with uncertainty. *Journal of Clinical Nursing*. 2017;26(23-24):4390-4403. doi:https://doi.org/10.1111/jocn.13768
26. Branchett K, Stretton J. Neonatal palliative and end of life care: What parents want from professionals. *Journal of Neonatal Nursing*. 2012;18(2):40-44. doi:10.1016/j.jnn.2012.01.009
27. Brashers DE. Communication and Uncertainty Management. *Journal of Communication*. 2001;51(3):477-497. doi:10.1111/j.1460-2466.2001.tb02892.x
28. Epstein RM, Street Jr. RL. Patient-centered communication in cancer care: Promoting healing and reducing suffering. Published online 2007. doi:10.1037/e481972008-001
29. Politi MC, Han PKJ, Col NF. Communicating the Uncertainty of Harms and Benefits of Medical Interventions. *Med Decis Making*. 2007;27(5):681-695. doi:10.1177/0272989X07307270
30. Kalke K, Studd H, Scherr CL. The communication of uncertainty in health: A scoping review. *Patient Education and Counseling*. Published online February 1, 2021. doi:10.1016/j.pec.2021.01.034
31. Krick JA, Weiss EM, Snyder A, Haldar S, Campelia GD, Opel DJ. Living with the Unknown: A Qualitative Study of Parental Experience of Prognostic Uncertainty in the Neonatal Intensive Care Unit. *Am J Perinatol*. Published online January 3, 2020. doi:10.1055/s-0039-3402722
32. Akkermans AA, Lamerichs JMWJJ, Schultz MJM, et al. How doctors actually (do not) involve families in decisions to continue or discontinue life-sustaining treatment in neonatal, pediatric, and adult intensive care: A qualitative study. *Palliat Med*. 2021;35(10):1865-1877. doi:10.1177/02692163211028079
33. VERBI Software. *MAXQDA Plus 2020*.; 2020. Accessed March 13, 2021. <https://www.maxqda.com/help-mx20/welcome>

34. Silver C, Lewins A. *Using Software in Qualitative Research: A Step-by-Step Guide*. SAGE; 2014.
35. Miles MB, Huberman AM. Qualitative data analysis: A sourcebook of new methods. *Qualitative data analysis: a sourcebook of new methods*. Published online 1984:263-263.
36. Morse JM. The Significance of Saturation. *Qual Health Res*. 1995;5(2):147-149. doi:10.1177/104973239500500201
37. Masso M, Allingham SF, Banfield M, et al. Palliative Care Phase: Inter-rater reliability and acceptability in a national study. *Palliat Med*. 2015;29(1):22-30. doi:10.1177/0269216314551814
38. Mather H, Guo P, Firth A, et al. Phase of Illness in palliative care: Cross-sectional analysis of clinical data from community, hospital and hospice patients. *Palliat Med*. 2018;32(2):404-412. doi:10.1177/0269216317727157
39. Medendorp NM, Hillen MA, van Maarschalkerweerd PEA, et al. "We don't know for sure": Discussion of uncertainty concerning multigene panel testing during initial cancer genetic consultations. *Fam Cancer*. 2020;19(1):65-76. doi:10.1007/s10689-019-00154-4
40. Anderson RJ, Stone PC, Low JTS, Bloch S. Managing uncertainty and references to time in prognostic conversations with family members at the end of life: A conversation analytic study. *Palliat Med*. 2020;34(7):896-905. doi:10.1177/0269216320910934
41. Bhise V, Meyer AND, Menon S, et al. Patient perspectives on how physicians communicate diagnostic uncertainty: An experimental vignette study†. *International Journal for Quality in Health Care*. 2018;30(1):2-8. doi:10.1093/intqhc/mzx170
42. Gordon GH, Joos SK, Byrne J. Physician expressions of uncertainty during patient encounters. *Patient Educ Couns*. 2000;40(1):59-65. doi:10.1016/s0738-3991(99)00069-5
43. Krawczyk M, Gallagher R. Communicating prognostic uncertainty in potential end-of-life contexts: Experiences of family members. *BMC Palliative Care*. 2016;15. doi:10.1186/s12904-016-0133-4
44. Medendorp NM, Hillen MA, Visser LNC, et al. A randomized experimental study to test the effects of discussing uncertainty during cancer genetic counseling: Different strategies, different outcomes? *European Journal of Human Genetics*. Published online January 12, 2021:1-11. doi:10.1038/s41431-020-00799-1
45. Santhosh L, Chou CL, Connor DM. Diagnostic uncertainty: From education to communication. *Diagnosis*. 2019;6(2):121-126. doi:10.1515/dx-2018-0088
46. Orzalesi M, Aite L. Communication with parents in neonatal intensive care. *The journal of maternal-fetal & neonatal medicine: The official journal of the European Association of Perinatal Medicine, the Federation of Asia and Oceania Perinatal Societies, the International Society of Perinatal Obstetricians*. 2011;24 Suppl 1:135-137. doi:10.3109/14767058.2011.607682
47. Lotz JD, Daxer M, Jox RJ, Borasio GD, Führer M. "Hope for the best, prepare for the worst": A qualitative interview study on parents' needs and fears in pediatric advance care planning. *Palliat Med*. 2017;31(8):764-771. doi:10.1177/0269216316679913
48. Latour JM, van Goudoever JB, Schuurman BE, et al. A qualitative study exploring the experiences of parents of children admitted to seven Dutch pediatric intensive care units. *Intensive Care Med*. 2011;37(2):319-325. doi:10.1007/s00134-010-2074-3
49. Clarke JN, Fletcher P. Communication Issues Faced by Parents Who have a Child Diagnosed with Cancer. *J Pediatr Oncol Nurs*. 2003;20(4):175-191. doi:10.1177/1043454203254040
50. Verberne LM, Fahner JC, Sondaal SFV, et al. Anticipating the future of the child and family in pediatric palliative care: A qualitative study into the perspectives of parents and healthcare professionals. *Eur J Pediatr*. 2021;180(3):949-957. doi:10.1007/s00431-020-03824-z

51. Contro N, Larson J, Scofield S, Sourkes B, Cohen H. Family Perspectives on the Quality of Pediatric Palliative Care. *Archives of Pediatrics & Adolescent Medicine*. 2002;156(1):14-19. doi:10.1001/archpedi.156.1.14
52. Wigert H, Dellenmark Blom M, Bry K. Parents' experiences of communication with neonatal intensive-care unit staff: An interview study. *BMC Pediatr*. 2014;14. doi:10.1186/s12887-014-0304-5
53. Zimmermann K, Bergstraesser E, Engberg S, et al. When parents face the death of their child: a nationwide cross-sectional survey of parental perspectives on their child's end-of life care. *BMC Palliat Care*. 2016;15:30. doi:10.1186/s12904-016-0098-3
54. Barry LC, Kasl SV, Prigerson HG. Psychiatric Disorders Among Bereaved Persons: The Role of Perceived Circumstances of Death and Preparedness for Death. *The American Journal of Geriatric Psychiatry*. 2002;10(4):447-457. doi:10.1097/00019442-200207000-00011
55. Lotz JD, Jox RJ, Borasio GD, Führer M. Pediatric advance care planning from the perspective of health care professionals: A qualitative interview study. *Palliat Med*. 2015;29(3):212-222. doi:10.1177/0269216314552091
56. Kaye EC, Stall M, Woods C, et al. Prognostic Communication Between Oncologists and Parents of Children With Advanced Cancer. *Pediatrics*. 2021;147(6):e2020044503. doi:10.1542/peds.2020-044503
57. Hill C, Knafl KA, Docherty S, Santacroce SJ. Parent Perceptions of the Impact of the PICU Environment on Delivery of Family-Centered Care. *Intensive Crit Care Nurs*. 2019;50:88-94. doi:10.1016/j.iccn.2018.07.007
58. Longden JV. Parental perceptions of end-of-life care on paediatric intensive care units: a literature review. *Nursing in Critical Care*. 2011;16(3):131-139. doi:10.1111/j.1478-5153.2011.00457.x
59. Evans LR, Boyd EA, Malvar G, et al. Surrogate Decision-Makers' Perspectives on Discussing Prognosis in the Face of Uncertainty. *Am J Respir Crit Care Med*. 2009;179(1):48-53. doi:10.1164/rccm.200806-969OC
60. van der Velden NCA, van der Kleij MBA, Lehmann V, et al. Communication about Prognosis during Patient-Initiated Second Opinion Consultations in Advanced Cancer Care: An Observational Qualitative Analysis. *Int J Environ Res Public Health*. 2021;18(11):5694. doi:10.3390/ijerph18115694
61. Freeman G. Progress with Relationship Continuity 2012, a British perspective. *International Journal of Integrated Care*. 2012;12(2). doi:10.5334/ijic.975
62. Bonney A, Phillipson L, Jones S, Iverson D. Older patients' attitudes to general practice registrars. A qualitative study. *Australian family physician*. 2009;38:927-931.
63. Tarrant C, Dixon-Woods M, Colman AM, Stokes T. Continuity and Trust in Primary Care: A Qualitative Study Informed by Game Theory. *Ann Fam Med*. 2010;8(5):440-446. doi:10.1370/afm.1160
64. Torke, A. M., Corbie-Smith, G. M., & Branch, W. T. (2004). African American patients' perspectives on medical decision making. *Archives of Internal Medicine*, 164(5), 525–530. <https://doi.org/10.1001/archinte.164.5.525>
65. Holloway RG, Gramling R, Kelly AG. Estimating and communicating prognosis in advanced neurologic disease. *Neurology*. 2013;80(8):764-772. doi:10.1212/WNL.0b013e318282509c
66. Srivastava R. Dealing with Uncertainty in a Time of Plenty. *N Engl J Med*. 2011;365(24):2252-2253. doi:10.1056/NEJMp1109456

67. Blanch-Hartigan D, van Eeden M, Verdam MGE, Han PKJ, Smets EMA, Hillen MA. Effects of communication about uncertainty and oncologist gender on the physician-patient relationship. *Patient Educ Couns*. 2019;102(9):1613-1620. doi:10.1016/j.pec.2019.05.002
68. Zhong L, Woo J, Steinhardt MA, Vangelisti AL. "Our Job is that Whole Gray Zone in Between There": Investigating Genetic Counselors' Strategies for Managing and Communicating Uncertainty. *Health Communication*. 2020;35(13):1583-1592. doi:10.1080/10410236.2019.1654171
69. Timmermans S, Yang A, Gardner M, et al. Does Patient-centered Care Change Genital Surgery Decisions? The Strategic Use of Clinical Uncertainty in Disorders of Sex Development Clinics. *J Health Soc Behav*. 2018;59(4):520-535. doi:10.1177/0022146518802460