Sharing the burden of deciding: How physicians and parents make end-of-life decisions
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Sharing the burden. How physicians and parents make end-of-life decisions.

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Sharing the burden
How physicians and parents make end-of-life decisions
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Who should decide?

This thesis started with a phone call somewhere in the autumn of 2005. At that time I had just returned to the Emma Children’s Hospital AMC to manage a hospital-wide program to improve the care for children with severe disabilities and their parents. On that day the mother of a 16 year old boy called me to share her story. This was what she told me about her son whom I will call Tom.

From birth on Tom had had severe cognitive, motoric and sensory disabilities. The cause was still unknown. Because of his disabilities he was not able to talk, to walk or to take care of himself. His eye sight was limited. Yet, he was able to laugh, to cry and to make small noises which his parents had learned to read. He recognized his parents and other close caregivers. Tom especially enjoyed listening to music and going out in his wheel chair in windy weather. Over the years, he had developed serious health problems. These included hard-to-treat epilepsy, recurring pneumonias and feeding problems. Because of these problems he had been given a PEG tube at the age of 10. In the last year, he had been admitted to the pediatric intensive care unit twice because of serious breathing problems. Both times he had needed mechanical ventilation in combination with intravenous antibiotics. The second time it had been difficult to wean him from ventilation, but he finally managed to breathe on his own again. After four weeks on the intensive care, he was able to return home. He remained too weak to go to the day care center. Yet, most of the days he seemed comfortable and reacted with a smile when he saw his parents or heard his favorite music. On an evening just a week ago, Tom got serious seizures which developed into a status epilepticus. Despite repeated doses of Stesolid he did not regain consciousness. The parents phoned their general practitioner. He advised
them to go immediately to the first aid unit. Still unconscious, Tom was seen by the attending pediatrician whom they had never met before. After having looked at Tom, he said a transfer to the pediatric intensive care unit might be necessary. He would phone the attending pediatric intensivist for further advice. Somewhat later he returned. He reported that the intensivist had expressed his strong doubts whether Tom should be admitted again to the intensive care unit. In fact, he rather wished to protect Tom from a next long, burdensome and probably complicated stay and focus on comfort care, either on the general ward or at home. At that moment, Tom started to regain consciousness. A few hours later his parents could take him home. Over the week they started to feel angry as well as confused. Could doctors decide on their own that Tom would not be admitted again to the intensive care unit? And on what grounds? Should not the parents make this decision because they knew their child best? If Tom would not have been severely disabled, wouldn’t he be immediately admitted and fully treated? To what extent had this doctor’s opinion about quality of life influenced his decision-making? And had an intensivist a greater say in decisions to withhold or withdraw treatment than a pediatrician?

These were the questions Tom’s mother hoped I could answer. And I told her I could not. That same day I asked the head of the pediatric intensive care unit whether he would speak with Tom’s parents about a future treatment plan for their son. And I asked him if we should not start doing more research into these questions. He answered that he would and that we should. And so I began to write a research plan which over the years evolved into this thesis.

**Treatment decisions**

The installment of any form of treatment will always be preceded by the question whether it will truly help the patient. In other words: will it be effective to solve or to alleviate the health problems for which it is installed and will it be proportional, i.e. will the expected benefits outweigh the expected harms. Often, these questions can be answered with a clear yes. In that case, the physician will inform the patient about what will be done and why, the patient will ask some clarifying questions, and after having given his consent the treatment will be started or a surgery will be scheduled. Occasionally, the questions whether a form of treatment will be effective and proportional has to be answered with a no. In that case, physician and patient will extensively discuss the alternative options and their pros and cons. But if none of these options are considered to be appropriate, physician and patient may decide to ‘wait and see’ or in case of a life-threatening illness may decide to focus on optimal palliative care. Sometimes, the question whether a course of treatment will truly help the patient cannot be answered with a clear yes or no. It may be that because of the patient’s co-morbidity it is hard to predict whether treatment will have any effect. It may even be that the patient will come out of this treatment in a worse state. Moreover, there may be a chance that the negative side effects may become too great a burden compared to the expected outcomes. In these cases, a simple exchange of information is not sufficient. Instead, the physician and the patient will need time to extensively discuss the different scenarios, the patient’s values in life and their mutual preferences. Based on this exchange, they can reach a decision together on what is best to do for this patient in this situation. Although many adult patients favor this approach of shared decision-making, not all of them do. There is evidence that especially in such uncertain situations some patients wish to be informed and to express their wishes, but they want to leave the final decision to their physician.¹³⁻⁴

**Children’s say in treatment decisions**

How children wish to be involved in decisions concerning their treatment is still largely unknown. The scarce studies which have been performed focus on children between 8 and 18 years of age, mainly diagnosed with cancer. What we know from these studies, is that most children favor to be honestly informed about their ongoing treatment and its effects in a language they can understand by a physician who is genuinely interested in them and has sufficient time. Children who have been informed in this way report that this helps them to feel better prepared and less anxious.⁴⁻⁸ Several children underline that they wish to be informed together with their parents. In their opinion, their parents are their main advocates and interpreters.⁴⁻⁸ While all children wish to be well informed and to express their questions, not all children wish to share in making decisions about their course of treatment. Some children wish to leave the decision-making to their physician and parents, especially if complex decisions have to be made. Other children wish to participate in making crucial decisions or even have the final say.⁴⁻⁸ In one study focusing on end-of-life care most children being included, aged between 10 and 20 years, seemed well aware of the far-reaching consequences of their decisions. Main reasons why they wished treatment to be forgone were that they did not believe they could be cured.
and disabled children.\textsuperscript{12-15} As a result, the management of these symptoms has substantially improved. A relevant issue for further discussion is the weight being given to non-verbal signals in decision-making. For example, parents as well as nurses sometimes observe that a premature baby starts shrinking away from every touch or becomes more and more apathetic. Parents of severely disabled children sometimes see that their child starts to persistently pull out his PEG tube or IV line or starts to vehemently resist dialysis. Interpreting these signals as ‘non-verbal wishes’ may go a step too far, but they may well be a sign of how burdensome treatment has become. Consequently, the observations of parents and other caregivers of these signals should be seriously weighed in decisions about whether treatment should be continued or forgone.

**Parental involvement**

The days are gone that the medical team took over all care as soon as a child was admitted to the hospital. In most countries, parents are now allowed to visit their sick child as long as they wish and to actively participate in his or her daily care. They also have the right to read their child’s medical record and to be informed timely about which tests will be done and which treatment will be given why, when and how. In medical centers in several countries, parents’ involvement stops here. Parents are informed, but they are not in any way included in the decision-making process regarding which medical treatments should be installed, continued or forgone. This paternalistic approach seems largely motivated by the physician’s wish to not burden parents with difficult decisions for which they lack the necessary medical knowledge.

In other countries, a different approach is recommended which may best be characterized as semi-shared. Here, parents are invited to express their viewpoints and wishes regarding the course of their child’s medical record and to be informed timely about which tests will be done and which treatment will be given why, when and how. In medical centers in several countries, parents’ involvement stops here. Parents are informed, but they are not in any way included in the decision-making process regarding which medical treatments should be installed, continued or forgone. This paternalistic approach seems largely motivated by the physician’s wish to not burden parents with difficult decisions for which they lack the necessary medical knowledge.\textsuperscript{16}

Recently, a tool has been developed to assess on an individual basis whether a child is able to give his or her well-balanced consent to participate in research.\textsuperscript{11} Outside of a research context, this tool might also be helpful to assess whether a child below 12 years of age is mature enough, cognitively, socially as well as emotionally, to be involved in making crucial treatment decisions. This could also apply to the decision for euthanasia. In the Netherlands, children between the age of 12 and 16 are allowed to make this decision under the same strict conditions as adults and with the additional criterion that the child’s parents agree. The Dutch Pediatric Society (NVK) has recently started a debate concerning the question whether the age for euthanasia should be decreased, following the recent change in the Belgian legislation.

While children from about the age of two are already able to give some words to how they experience and cope with their medical treatment, younger children as well as children with serious cognitive disabilities can only express themselves by means of non-verbal communication. Over the last ten years, the assessment of non-verbal signals of pain and other symptoms of discomfort has received considerable attention in neonates and disabled children.\textsuperscript{12-15} As a result, the management of these symptoms has substantially improved. A relevant issue for further discussion is the weight being given to non-verbal signals in decision-making. For example, parents as well as nurses sometimes observe that a premature baby starts shrinking away from every touch or becomes more and more apathetic. Parents of severely disabled children sometimes see that their child starts to persistently pull out his PEG tube or IV line or starts to vehemently resist dialysis. Interpreting these signals as ‘non-verbal wishes’ may go a step too far, but they may well be a sign of how burdensome treatment has become. Consequently, the observations of parents and other caregivers of these signals should be seriously weighed in decisions about whether treatment should be continued or forgone.

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Finally, there are countries like the United States and Canada where parents are considered to be the final decision-makers for their child, also regarding decisions to withhold or withdraw life-sustaining treatment. Yet, this ‘premise’ that parents bear final responsibility may lead to different approaches in clinical practice. It may either result in an informed approach in which parents make the decision alone, based on all information being provided to them, or in a shared approach in which parents decide in close cooperation with their child’s physician.\(^{18-21}\)

An approach which one couple of parents finds helpful in coping with difficult decisions may not be favored by another couple of parents. It may also be that the preferences of both parents or of one of the parents change over time. For instance, parents may wish to fully share in the decision to not resuscitate their child again in future. But after their child has been acutely admitted to the intensive care unit, they may wish to let their physician decide about if and when mechanical ventilation shall be withdrawn. There is growing consensus that physicians should tailor their decision-making approach to parents’ preferences.\(^{18, 20-30}\) It is also recommended that they tailor their communication to parents’ needs.\(^{31-34}\) But how can physicians best do this? Should they ask parents directly to what extent they wish to be involved in the decision-making process? How extensively should they inform them about all concerns and remaining options? Can parents be asked what they think is best to do? Will they not always favor treatment to be continued? To answer these questions and formulate valid recommendations, more knowledge is needed about how treatment decisions are communicated with parents in actual practice. Yet, prospective observational studies are scarce and the few studies which have been done do not provide a detailed analysis of the communication behaviors of the physicians and the parents.

Although decisions to withhold or withdraw treatment are considered to be part of ‘normal medical practice’ in most Western countries, reaching this decision is never felt as normal practice. As physicians often underline, it is a decision you can only take once, with the child’s death as the most probable and sometimes certain outcome. Even if parents and medical team fully agree about its inevitability and share the conviction that the child’s suffering should not be prolonged any longer, this decision will cause strong emotions in parents and often in the professionals involved as well. This process becomes even more difficult if parents and the medical team start to have divergent points of view on what is in the child’s best interests to do. It may be presumed that parents will more often wish treatment to be continued while according to the medical team this is no longer appropriate. The scarce studies exploring conflicts in end-of-life decision-making point in this direction.\(^{36, 37}\) They also indicate that conflicts which have not been handled well may complicate parents’ mourning in the long term.\(^{38}\) This necessitates further research on the main causes of conflict and on the strategies which physicians and parents apply to resolve these conflicts.

Death rates in relation to end-of-life decisions in children

According to recent information provided by UNICEF, 17,000 children between 0 and 6 years of age die worldwide each day. The vast majority of children die from malnutrition and infections, due to extreme poverty, unsafe water, poor hygiene and lacking basic health care. Hence, most children die in the least developed, conflict-affected countries. Their deaths are not preceded by deliberate decisions to withhold or withdraw life-sustaining treatment. The only decision parents might feel forced to make is to not seek treatment if their child becomes critically ill. First, because they do not have the financial means to pay for this treatment and second, because they cannot afford to care for a severely disabled child in case their child would survive. In a recent paper ‘My job is to keep him alive, but what about his brother and sister?’ Indian doctors reported that lack of resources, usually ventilators, forced them to make decisions about which babies should get the chance to live. Moreover, they were well aware that their decision to continue or discontinue a baby’s treatment directly influenced the family’s economy and reputation as well as the availability of food and education for siblings. This made them feel even more responsible and often caused moral distress.\(^{37}\)

These facts stand in sharp contrast with the situation in rich countries like the Netherlands. Over the last 100 years, the mortality rate in Dutch infants and children decreased to 3.8 of 1000 infants between 0 and 1 years of age and to 1 of 10,000 children between 1 and 16 years of age. In 2012, approximately 600 infants died, mainly from prematurity or congenital disorders. In that same year, approximately 400 children between 1 and 16 years of age died.\(^{37, 38}\) Main causes were trauma and cancer. Some children could not be reached in time by an ambulance. But the majority of children received highly specialized care prior to their death. This confronted both physicians and parents with a very different dilemma: should treatment be continued at all costs to prolong the child’s life as long as possible? Or would
it be more humane to forgo treatment at some point to allow the child to
die peacefully, preferably at home? There is international consensus that
life-prolonging treatment may be withheld of withdrawn when all active
treatment options have been exhausted and a child has no chance of survival.
Or, to state it more precisely, when the child’s death seems both inevitable
and imminent. At the same time, there is still a great deal of dissent about
whether a very poor quality of life may be a decisive reason to forgo life-
sustaining treatment in children who cannot speak for themselves. The
international controversy is even greater when this decision is based on
the predicted quality of life. Yet, in the Netherlands physicians largely
agree that the actual and the predicted quality of life should be taken into
account in end-of-life decisions in children. Currently, 68% of all deaths in
Dutch children between 0 and 1 year of age are now preceded by a decision
to discontinue treatment. In children between the ages of 1 and 17 years, this
rate is 36%.41

**Personal emotions and personal values**

While end-of-life decision-making will cause emotional reactions,
in the parents as well as in professionals directly involved in the child’s care,
these emotions in turn will influence the decisions being made. Parents’
intense fear of losing their child may increase their wish to postpone a
decision to withhold or withdraw treatment as long as possible. But their
grief about their child’s prolonged suffering may eventually make them
wish that their child will be allowed to die. There are conflicting opinions
about whether physicians should strive towards making rational, ‘emotion-
free’ decisions and, as important, if they will succeed in doing so. A recent
study among Australian and New Zealand neonatologists showed that their
emotions significantly influenced their attitudes. Physicians with a greater
fear of the dying process and a greater fear of premature death were more
likely to accept hastening a newborn infant’s death with analgesia-sedation.42

Besides emotions, personal values and beliefs appear to have a
significant influence on end-of-life decision-making. For instance, in a
recent study among Israeli physicians, very religious physicians, compared
to moderately religious and secular physicians, were less likely to believe
that life-sustaining treatment should be withdrawn. It may be presumed
that very religious parents share this conviction. A survey among European
neonatologists also illustrated the significant influence of physicians’
personal values. Neonatologists from Eastern and Southern European
countries, mostly with a Catholic background, largely agreed that life should
be preserved at any cost, while neonatologists from Northern European
countries, mostly with a Protestant or a secular background, underlined that
quality-of-life arguments should be taken into account. According to both
groups of physicians, their views directly influenced their clinical decision-
making. The following outcome was of even more interest. The majority of
physicians in most countries reported that the presence of mental rather
than physical disability and the burden to the family were factors they
considered to be important in making decisions to limit intensive neonatal
interventions.39

When physicians and parents share the same personal values and
beliefs, this may add to a concordant decision-making process which in turn
may be considered helpful for the parents as well as their physicians. But will
it always be in the best interests of the child? Let me sketch two scenarios
to which it may lead. On the one extreme, a scenario could evolve in which
both parents and physicians agree that the child’s life should be prolonged at
any cost, resulting in continued treatment with prolonged suffering until the
child dies under full support. Or resulting in survival of the child for the time
being but without any ability to interact and communicate, because of very
severe impairments, and with considerable discomfort. On the other extreme,
a scenario could evolve in which both parents and physicians agree that only
if a child has the cognitive abilities to eventually lead an independent life
this may be called a life ‘worth living’. This may result in the withholding
of medical care which would normally be given to a child, and eventually in
the child’s death as irreversible outcome. Both scenarios illustrate that a
harmonious decision-making process in itself does not guarantee that the
child’s interests are being guarded in the best possible way.44 At the same
time, they illustrate the complexity of making end-of-life decisions for a
child. For instance, in the second scenario parents’ decision may not have
been influenced by personal values, but by their fear of not being able to
take care of a severely disabled child. This complexity underlines the need
of combining empirical research into how physicians and parents make
decisions in clinical practice with ethical analyses of how decisions should be
made in the best interests of all involved, the child first of all.
Thesis Outline

This thesis consists of three parts. In the first part, we focus on the doctors’ perspective and investigate how pediatric specialists make end-of-life decisions, how they involve the child and his or her parents in the decision-making process, and how they cope with conflicts. In the second part, our main focus are the parents. We explore how parents from different countries experience their involvement in end-of-life decisions for their children and which factors they consider to be important in making these decisions. In the third part, entitled the shared perspective, we focus on both the perspective of the doctors and of the parents. We investigate how physicians and parents communicate about end-of-life decisions in clinical practice and to what extent parents participate in making these decisions. Moreover, we explore the pros and cons of making decisions together with parents and based on this analysis formulate recommendations how to better involve and support parents throughout the complex process of end-of-life decision-making.

In chapter 2 we present the outcomes of a cross-sectional national survey among pediatric specialists who are regularly involved in making end-of-life decisions in children and who are working in one of the eight Dutch university hospitals. For this aim we developed a questionnaire, partly based on previous Dutch studies on end-of-life decision-making in neonatology. Of the 185 eligible pediatric specialists, 136 returned the questionnaire.

In chapter 3 we present the outcomes of a national survey among Dutch neonatologists, working in one of the 10 Dutch neonatal intensive care units. We interviewed the 85 attending physicians of 147 newborns who died between December 2006 and October 2007 after an end-of-life decision had been made on the basis of the child’s poor prognosis.

In chapter 4 we present the outcomes of an integrative review. We searched PubMed, EMBASE, CINAHL and PsycInfo for original studies published between January 2000 and July 2010 which had directly investigated the experiences of parents of neonates and children for whom a decision to withhold or withdraw LST had been made. We identified 17 studies that met all inclusion criteria.

In chapter 5 we present the outcomes of a prospective exploratory study on the PICUs of two Dutch University Medical Centers. The first inclusion period lasted from April 2008 until April 2009, the second from April 2010 until April 2011. Overall, 27 physicians participated, along with 37 parents of 19 children for whom an end-of-life decision was being considered. Forty-seven conversations were audio recorded, ranging from 1 to 8 meetings per patient. By means of a coding instrument, we quantitatively and qualitatively analyzed physicians’ and parents’ communication behaviors.

In chapter 6 we present the outcomes of an ethical analysis. For this purpose, we first describe the decision-making process for a 3-year-old boy with severe acquired brain damage where the parents wanted to stop further treatment contrary to the views of the medical team. By comparing this case with relevant literature, we systematically explore the pros and cons of sharing end-of-life decisions with parents in cases where treatment is considered futile by parents and not (yet) by physicians. We collected data by chart review, by audio recordings of team discussions and meetings with the parents, and by in-depth interviews with the parents and the primary care professionals within 6 months after the patient’s death. We wrote this chapter together with the patient’s parents.

Finally, in chapter 7 we present the overall conclusions and discuss the main strengths and limitations of this research project. We will close this chapter with recommendations to improve a shared process of end-of-life decision-making in children.
# End-of-life decisions

Decisions that may unintentionally or intentionally hasten death, including withholding or withdrawing medical treatment.

## Normal medical practice

- Withdrawing or withholding futile treatment;
- Withdrawing or withholding treatment at the patient’s request (in patients aged 12 years and older);
- Administering medication to alleviate pain (or other forms of serious suffering);
- Palliative sedation.

## Palliative sedation

Administration of medication aimed at lowering the patient’s consciousness to alleviate pain and suffering at the end of life. It is permissible when a patient’s life expectancy is no more than 2 weeks.

## Euthanasia

Since 2002, euthanasia is allowed if certain conditions are met. A regional review committee will assess whether the following due care criteria have been met:

- The patient’s suffering is unbearable and with no prospect of improvement;
- The patient’s request for euthanasia must be voluntary, competent and persistent over time;
- The patient must be fully aware of his/her condition, prospects and options;
- There must be consultation with at least one other independent physician who needs to confirm the conditions mentioned above;
- Euthanasia must be carried out in a medically responsible and careful manner by the physician;
- The patient is at least 12 years old (patients between 12 and 16 years of age require the consent of their parents).

## Groningen Protocol

Under very strict conditions Dutch case law allows physicians to deliberately end a newborn’s life (the so-called ‘defense of necessity’). These conditions are further elaborated in the Groningen Protocol, published in 2004. Each of the following conditions has to be properly fulfilled:

- The child’s suffering must be unbearable and with no prospect of improvement;
- Both the physician and parents must be convinced that there is no reasonable alternative solution given the child’s situation;
- The parents must have given their agreement for the termination of life;
- There must be no doubt about the diagnosis and prognosis;
- At least one other independent physician must have examined the child and given a written opinion on compliance with the due care criteria listed above;
- The termination must be performed with all due care; and must be reported to the local medical examiner, the district attorney and a national review committee.

## Consent of the child

Patients aged 16 years and older have to give their consent for each separate action by a medical professional. In children between 12 and 16 years of age the consent of the child and the agreement of legal representative(s), mostly the parents, are needed.

## Agreement of parents

In children between 0 and 12 years, the physician’s liabilities are met towards the legal representative(s), mostly the parents. The same applies to children aged 12 years and older when they are not capable of reasonably advocating their own interests.

## Responsibilities of physicians

Physicians bear final responsibility for the decision that treatment has become futile, based on the latest professional standards and knowledge. Parental agreement is not required. Parents cannot oblige physicians to start or continue a futile treatment.

### TABLE 1  Legal context regarding end-of-life decision-making in the Netherlands

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At some point you know that you can no longer avoid the subject. Either because you see that the child’s death is imminent. Or because the child is suffering to such an extent that I think our treatments are not doing any good anymore. Ideally, you decide together with parents. With each of us having different roles. The doctor with some objective distance and the parents knowing their child deeply. In most cases, you are able to work it out together, as long as there is no time pressure.

Quote from interview with one of the participating physicians
The process of end-of-life decision-making in pediatrics: a national survey in the Netherlands

Chapter 2

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Albert P. Bos

Pediatrics 2011;127:e1004-e1012
Abstract

What’s known on this subject: Studies within neonatology and pediatric critical care have revealed that physicians consider it important to share end-of-life decisions with parents and to take sufficient time to resolve conflicts. How shared decision-making evolves in everyday practice is not yet known.

What this study adds: Within Dutch pediatrics, end-of-life decisions are team decisions. The level of parental involvement varies greatly depending on type of decision and type and duration of treatment. Conflicts within the medical team and between parents and team occur regularly.

Objective: The goal of this study was to investigate how Dutch pediatric specialists reach end-of-life decisions, how they involve parents, and how they address conflicts.

Methods: We conducted a national cross-sectional survey among pediatric intensivists, oncologists, neurologists, neurosurgeons, and metabolic pediatricians practicing in the 8 Dutch university hospitals. We collected information on respondents’ overall opinions and their clinical practice.

Results: Of the 185 eligible pediatric specialists, 74% returned the questionnaire. All responding physicians generally discuss an end-of-life decision with colleagues before discussing it with parents. In half of the reported cases, respondents informed parents about the intended decision and asked their permission. In one-quarter of the cases, respondents informed parents without asking for their permission. In the remaining one-quarter of the cases, respondents advised parents and consequently allowed them to have the decisive voice. The chosen approach is highly influenced by type of decision and type and duration of treatment. Conflicts within medical teams arose as a result of uncertainties about prognosis and treatment options. Most conflicts with parents arose because parents had a more positive view of the prognosis or had religious objections to treatment discontinuation. All conflicts were eventually resolved by a combination of strategies. In 66% of all cases, pain and symptom management were intensified before the child’s death.

Conclusions: Within Dutch pediatrics, end-of-life decisions are team decisions. Pediatric specialists differ considerably in how they involve parents in end-of-life decision-making, ranging from benevolent paternalism to parental autonomy. Main conflict-solving strategies are taking more time and extending discussions.

2.1 Introduction

One hundred years ago, 1 of 10 children in the Netherlands died before his or her first birthday. Because of improved socioeconomic circumstances and the increase in medical-technical possibilities, the mortality rate in Dutch infants and children decreased to 3.8 of 1000 infants between 0 and 1 year of age and 1 of 10 000 children between 1 and 16 years of age in 2008. Congenital disorders are the main cause of death in infants, whereas trauma and malignancies are the main causes of death in older children. The rapid increase in medical-technical possibilities has raised the dilemma whether everything that is technically possible should always be applied. Currently, the common opinion among Dutch pediatricians is that life should not be prolonged at any cost. This is reflected by the fact that 68% of all deaths in Dutch children between 0 and 1 year of age are preceded by a decision to discontinue treatment. In children between 1 and 16 years of age, this rate is 36%.

In 2005, the so-called Groningen protocol defined that under very strict conditions the use of sedatives and analgesics with the intention to hasten death can be acceptable for neonates with extensive anomalies and unbearable and untreated suffering. Since then, the Netherlands has been labeled as (and criticized for being) a country with liberal practices concerning end-of-life decision-making (EoLDM). To date, Dutch research on EoLDM has focused on neonatology. How this decision-making process takes place in infants and children has not yet been explored. Of special interest is the position of parents in this process. International guidelines on EoLDM give contradictory recommendations regarding their level of involvement and authority.
For the present study, we conducted a cross-sectional national survey among Dutch pediatric specialties involved in the process of EoLDM in children. Our main questions were (1) how do pediatric specialists reach end-of-life decisions, and how do they involve parents in this process? and (2) how are conflicts within the medical team and between parents and the team addressed? This survey is the first phase of a research program entitled “Tailor Made Decision Making Within Dutch Pediatrics.” The purpose of this program is to collect more scientific knowledge of EoLDM in infants and children as well as to develop guidelines on how to better guide parents throughout this process. It is funded by the Netherlands Organisation for Health Research and Development.

2.2 Methods

Study design

We conducted a national cross-sectional survey among medical specialists in the Netherlands involved in end-of-life care in children between 0 and 16 years of age.

Study population

In the Netherlands, children with life-threatening illnesses are mostly referred to 1 of the 8 university hospitals. All staff members and fellows of the departments of pediatric intensive care, oncology, neurology, neurosurgery, and metabolic diseases of these 8 university hospitals (n = 185) received an anonymous questionnaire. Because our study did not focus on neonates, neonatologists were not included. The survey was conducted between February 2008 and January 2009.

Survey questionnaire

A detailed questionnaire, partly based on previous Dutch studies on EoLDM within neonatology, was developed. This questionnaire consisted of 43 precoded and 2 open questions, divided into 3 sections. It took respondents ~ 45 minutes to complete. The first section gathered data on respondent characteristics such as age, gender, medical specialty, and religion. The second section contained 10 questions, assessing general opinions and practices concerning EoLDM. Twenty-five questions in the third section referred to the most recent case of a child who had been treated by the respondent and had met the following criteria: (1) the child had died before the age of 16 years; and (2) a decision to withhold or withdraw medical treatment had preceded death. The definitions used in our study are listed in Table 1.
2.3 Results

Respondent characteristics

Of the 185 eligible pediatric specialists, 136 returned the questionnaire (74%). Table 2 summarizes the main characteristics of the respondents. Their mean age was 44 years.

<table>
<thead>
<tr>
<th>TABLE 2</th>
<th>Characteristics of pediatric specialists</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Intensivists</td>
</tr>
<tr>
<td></td>
<td>(N=59)</td>
</tr>
<tr>
<td>Gender, male</td>
<td>28 (48)</td>
</tr>
<tr>
<td>Experience</td>
<td></td>
</tr>
<tr>
<td>0-5 y</td>
<td>26 (46)</td>
</tr>
<tr>
<td>5-10 y</td>
<td>10 (17)</td>
</tr>
<tr>
<td>&gt;10 y</td>
<td>21 (37)</td>
</tr>
<tr>
<td>Religion</td>
<td></td>
</tr>
<tr>
<td>Christian</td>
<td>16 (28)</td>
</tr>
<tr>
<td>Islamic</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Other</td>
<td>3 (5)</td>
</tr>
<tr>
<td>None</td>
<td>39 (67)</td>
</tr>
<tr>
<td>Diagnosis in most recent case</td>
<td></td>
</tr>
<tr>
<td>Congenital disorder</td>
<td>37 (70)</td>
</tr>
<tr>
<td>Cancer</td>
<td>6 (11)</td>
</tr>
<tr>
<td>Acute illness</td>
<td>10 (19)</td>
</tr>
</tbody>
</table>

Data analysis

Statistical analyses were performed in SPSS 16.0 (SPSS Inc, Chicago, IL). We used descriptive statistics to assess characteristics of the medical specialists and the patients who were presented as most recent cases. To analyze the influence of physician, patient, and family characteristics on the decision-making process and the arising of conflicts, \( \chi^2 \) tests were performed. If possible, overall group differences were tested before differences between specific categories were tested. A significance level of .05 was used for all analyses.

* Because of missing information, not all columns total N.
* We combined neurosurgeons and neurologists in all statistical analyses, because of the small number of neurosurgeons and the similarities in background and information compared with the neurologists.
* Defined as the most recent case of a child between 0 and 16 years of age who was treated by the respondent and who died after a decision to withhold or withdraw medical treatment.
General opinions and practices

All respondents reported that the decision to withhold or withdraw life-prolonging treatment is almost always discussed within the medical team before it is discussed with the parents. According to 40% of respondents (54 of 136), the final responsibility for an end-of-life decision lies with the medical team; according to 39% (53 of 136), with the physician in charge; and according to 21% (29 of 136, mostly oncologists), with both the physician in charge and the parents.

The majority of respondents prefer to raise the subject of how to proceed with life-sustaining treatment before acute life-threatening problems arise (59% [73 of 123]). Twenty-five percent of the respondents (34 of 136) reported the use of a locally developed guideline. Development of a national practice guideline on EoLDM in children would be welcomed by 44% of the respondents (60 of 136).

Most recent cases

Eighty-five percent of the respondents (116 of 136) completed the third section of the questionnaire about the decision-making process in their most recent case. The remaining respondents gave 2 reasons for not completing this section: lack of time or no eligible case in the last 5 years.

Case characteristics

Clinical characteristics of the presented cases are listed in Table 3. The majority of the children had a congenital disorder (55% [64 of 116]), followed by children with cancer (33% [38 of 116]) and children with an acute illness, including neurotrauma, acute infection, and asphyxia (12% [14 of 116]). Most of the children with congenital disorders (83% [53 of 64]) and with acute illnesses (86% [12 of 14]) received life-sustaining therapy (LST), consisting of mechanical ventilation. Children with cancer relatively often received comfort care without LST (47% [18 of 38]). At the time of the decision-making, the majority of children with congenital disorders or an acute illness either had a very limited life expectancy of less than a week or an unclear prognosis. Children with cancer often had a somewhat longer life expectancy but without a chance of survival.

Type of decision and implementation

In the end, treatment was withheld in 44% of the patients (51 of 116) and withdrawn in 56% (65 of 116). The decision to withhold treatment included withholding mechanical ventilation (51% [26 of 51]), chemotherapy (20% [10 of 51]), resuscitation (18% [9 of 51]), surgery (16% [8 of 51]), and/or antibiotics (8% [4 of 51]). The decision to withdraw treatment included withdrawal of mechanical ventilation (74% [48 of 65]), medication (11% [7 of 65]), chemotherapy (8% [5 of 65]), surgery (2% [1 of 65]), parenteral feeding (2% [1 of 65]), and/or extracorporeal membrane oxygenation (2% [1 of 65]).

Pain and symptom relief were intensified in 66% (76 of 116) of all patients. In 13% of these patients (10 of 76), hastening of death was intended besides alleviation of pain and other symptoms. If treatment was withdrawn, children died significantly more often within the first 24 hours than if treatment was withheld (60% vs 28%; $\chi^2 = 12.3$, P <.001).

Discussing end-of-life decisions

Discussions about withholding or withdrawing treatment were initiated by the medical team in 75% of the cases (87 of 116), by the parents, and in 21% (24 of 116) by both. In 49% of the cases (56 of 115 [in 1 case, information on parental involvement was missing]), the intended decision was presented as a medical decision for which parental permission was sought. In 24% of the cases (28 of 115), the decision was presented as a medical decision without explicitly asking for parental permission. Reasons for not asking permission were: the decision to withhold or withdraw life-prolonging treatment was regarded as a medical issue; respondents did not want to burden parents with this question; respondents were convinced parents would agree on the basis of former discussions. In 27% (31 of 115) of the cases, the decision was presented as medical advice, and parents were allowed to have the decisive voice.

As shown in Table 4, parents were most often allowed to have the decisive voice in cases of children receiving comfort care without LST. In contrast, in children receiving LST physicians significantly more often presented the decision to withhold or withdraw treatment as a medical decision, with or without asking parental permission, and not as medical advice. If withdrawal instead of withholding was considered, physicians also significantly more often presented this as a medical decision, thereby asking for parental permission, and not as medical advice (57% vs 37%; $\chi^2 = 4.4$, P =.035).
### TABLE 3 Case characteristics

<table>
<thead>
<tr>
<th></th>
<th>Congenital disorder (N=64)</th>
<th>Cancer (N=38)</th>
<th>Acute illness (N=14)</th>
<th>Total (N=116)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Patients</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-1 y</td>
<td>31 (48)</td>
<td>3 (8)</td>
<td>5 (36)</td>
<td>39 (34)</td>
</tr>
<tr>
<td>1-4 y</td>
<td>16 (25)</td>
<td>6 (16)</td>
<td>5 (36)</td>
<td>27 (23)</td>
</tr>
<tr>
<td>4-12 y</td>
<td>12 (19)</td>
<td>23 (60)</td>
<td>1 (7)</td>
<td>36 (31)</td>
</tr>
<tr>
<td>&gt;12 y</td>
<td>5 (8)</td>
<td>6 (16)</td>
<td>3 (23)</td>
<td>14 (12)</td>
</tr>
<tr>
<td>Mean age, y</td>
<td>3.2</td>
<td>7.4</td>
<td>4.4</td>
<td>4.7</td>
</tr>
<tr>
<td>Gender, male, n (%)</td>
<td>45 (70)</td>
<td>20 (53)</td>
<td>8 (57)</td>
<td>73 (63)</td>
</tr>
<tr>
<td>Duration of medical care, n (%)</td>
<td>14 (22)</td>
<td>3 (8)</td>
<td>10 (71)</td>
<td>27 (24)</td>
</tr>
<tr>
<td>1-6 mo</td>
<td>19 (30)</td>
<td>8 (22)</td>
<td>4 (29)</td>
<td>31 (27)</td>
</tr>
<tr>
<td>6-12 mo</td>
<td>10 (15)</td>
<td>5 (14)</td>
<td>0 (0)</td>
<td>15 (13)</td>
</tr>
<tr>
<td>&gt;1 y</td>
<td>21 (33)</td>
<td>20 (56)</td>
<td>0 (0)</td>
<td>41 (36)</td>
</tr>
<tr>
<td>Type of treatment(s) during EoLD, n (%)</td>
<td>25 (39)</td>
<td>15 (39)</td>
<td>3 (21)</td>
<td>43 (37)</td>
</tr>
<tr>
<td>Cure</td>
<td>53 (83)</td>
<td>20 (53)</td>
<td>12 (86)</td>
<td>85 (73)</td>
</tr>
<tr>
<td>Life-Sustaining Treatment</td>
<td>14 (22)</td>
<td>18 (47)</td>
<td>2 (14)</td>
<td>34 (29)</td>
</tr>
<tr>
<td>Life expectancy before EoLD, n (%)</td>
<td>32 (51)</td>
<td>12 (32)</td>
<td>9 (64)</td>
<td>53 (46)</td>
</tr>
<tr>
<td>&lt;1 wk</td>
<td>4 (6)</td>
<td>10 (26)</td>
<td>1 (7)</td>
<td>15 (13)</td>
</tr>
<tr>
<td>1 wk to 1 mo</td>
<td>12 (18)</td>
<td>12 (32)</td>
<td>0 (0)</td>
<td>23 (20)</td>
</tr>
<tr>
<td>&gt;1 mo</td>
<td>16 (25)</td>
<td>4 (10)</td>
<td>4 (29)</td>
<td>24 (21)</td>
</tr>
<tr>
<td>EoLD, n (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Withholding treatment</td>
<td>32 (50)</td>
<td>17 (45)</td>
<td>2 (14)</td>
<td>51 (44)</td>
</tr>
<tr>
<td>Withdrawing treatment</td>
<td>32 (50)</td>
<td>21 (55)</td>
<td>12 (86)</td>
<td>65 (56)</td>
</tr>
</tbody>
</table>

**Parents**

<table>
<thead>
<tr>
<th></th>
<th>Congenital disorder (N=64)</th>
<th>Cancer (N=38)</th>
<th>Acute illness (N=14)</th>
<th>Total (N=116)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intensifying pain and symptom management after withholding or withdrawing, n (%)</td>
<td>22 (35)</td>
<td>9 (24)</td>
<td>8 (57)</td>
<td>39 (34)</td>
</tr>
<tr>
<td>Yes</td>
<td>41 (65)</td>
<td>29 (76)</td>
<td>6 (43)</td>
<td>76 (66)</td>
</tr>
<tr>
<td>With intention to hasten death besides alleviation of pain and other symptoms</td>
<td>4 (10)</td>
<td>3 (10)</td>
<td>3 (50)</td>
<td>10 (13)</td>
</tr>
<tr>
<td>Interval between end-of-life decision and death, n (%)</td>
<td>28 (44)</td>
<td>12 (32)</td>
<td>12 (86)</td>
<td>52 (46)</td>
</tr>
<tr>
<td>&lt; 24 h</td>
<td>24 (38)</td>
<td>8 (22)</td>
<td>2 (14)</td>
<td>34 (30)</td>
</tr>
<tr>
<td>1-7 d</td>
<td>3 (5)</td>
<td>4 (11)</td>
<td>0 (0)</td>
<td>7 (6)</td>
</tr>
<tr>
<td>&gt; 1 mo</td>
<td>8 (13)</td>
<td>13 (35)</td>
<td>0 (0)</td>
<td>21 (18)</td>
</tr>
<tr>
<td>Marital status, n (%)</td>
<td>58 (91)</td>
<td>30 (79)</td>
<td>11 (79)</td>
<td>99 (85)</td>
</tr>
<tr>
<td>Married</td>
<td>3 (5)</td>
<td>7 (18)</td>
<td>2 (14)</td>
<td>12 (10)</td>
</tr>
<tr>
<td>Divorced</td>
<td>1 (1)</td>
<td>1 (3)</td>
<td>1 (7)</td>
<td>3 (3)</td>
</tr>
<tr>
<td>Other</td>
<td>2 (3)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>2 (2)</td>
</tr>
<tr>
<td>First language, n (%)</td>
<td>49 (78)</td>
<td>31 (82)</td>
<td>13 (93)</td>
<td>93 (81)</td>
</tr>
<tr>
<td>Dutch</td>
<td>14 (22)</td>
<td>7 (18)</td>
<td>1 (7)</td>
<td>22 (19)</td>
</tr>
<tr>
<td>Religious</td>
<td>23 (36)</td>
<td>17 (45)</td>
<td>6 (43)</td>
<td>46 (40)</td>
</tr>
<tr>
<td>Christian</td>
<td>14 (22)</td>
<td>5 (13)</td>
<td>0 (0)</td>
<td>19 (16)</td>
</tr>
<tr>
<td>Islamic</td>
<td>1 (2)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>1 (1)</td>
</tr>
<tr>
<td>Other</td>
<td>7 (11)</td>
<td>4 (10)</td>
<td>1 (7)</td>
<td>12 (10)</td>
</tr>
<tr>
<td>Unknown</td>
<td>19 (30)</td>
<td>12 (32)</td>
<td>7 (50)</td>
<td>38 (33)</td>
</tr>
</tbody>
</table>

EoLD indicates end-of-life decision.

* Because of missing information, not all columns total N.
* One or more answers were possible.
There was no significant correlation between pediatric specialty and the way a decision was presented to parents. Respondents working within the same specialty in the same center showed individual variability in how they presented a decision.

Conflicts within the medical team

In 13% of the cases (15 of 115), there was initial disagreement within the medical team about the intended decision to withhold or withdraw treatment. These disagreements were a result of conflicting views on the remaining treatment options (60% [9 of 15]), carefulness exercised in the decision-making process (40% [6 of 15]), life expectancy (27% [4 of 15]), expected quality of life (27% [4 of 15]), involvement of parents (13% [2 of 15]), or the moral justification of the decision (13% [2 of 15]). Conflicts were significantly more likely to arise in children with congenital disorders, compared with children with cancer or acute illnesses (20% vs 4%; $\chi^2 = 6.7$, $P = .032$). There were fewer conflicts about children receiving comfort care only, compared with children receiving LST or curative treatment (3% vs 18%; $\chi^2 = 4.5$, $P = .036$). Conflicts within the team were in all cases resolved by a combination of strategies: prolongation of team discussions (53% [8 of 15]), additional diagnostics (47% [7 of 15]), second opinions (33% [5 of 15]), or giving the medical director the decisive voice (13% [2 of 15]).

Conflicts between parents and the medical team

In 20% of the cases (23 of 116), parents and the medical team initially disagreed about the decision to withhold or withdraw treatment. In a majority of these 23 cases (57% [13 of 23]) there were ≥2 issues on which parents and team had conflicting points of view. Contrary to the opinion of the team, parents were convinced that there were still treatment options (43% [10 of 23]), believed that their child would survive (30% [7 of 23]), and/or had a more positive view on the expected quality of life of their child (35% [8 of 23]). These last 8 cases were all children with congenital disorders. In 6 cases, parents had strong moral objections against the intended end-of-life decision because of religious convictions (26% [6 of 23]).

As a result of these points of view, in 22 of the 23 cases parents wanted treatment to be continued. In the 1 remaining case, the parents thought that the decision-making process was going too slow, thereby prolonging their child’s suffering. They wished that treatment was withdrawn instead of withheld as proposed by the team. Parents with Dutch as their first language had fewer conflicts with the medical team compared with other parents (11% vs 50%; $\chi^2 = 18.6$, $P < .001$). Muslim parents were more often involved in conflicts with the medical team than non-Muslim parents (58% vs 12%; $\chi^2 = 22.1$, $P < .001$). Intrateam conflicts and conflicts between team and parents were highly interrelated ($\chi^2 = 17.3$, $P < .001$).

The 2 main strategies to address conflicts, often used in combination, were giving parents more time by delaying a decision (57% [13 of 23]) and intensifying discussions with parents (57% [13 of 23]). Other strategies were getting a second opinion (52% [12 of 23]), performing additional diagnostics (26% [6 of 23]), or taking over all responsibility for the decision-making process in agreement with parents (26% [6 of 23]). This mainly concerned Muslim parents (83% [5 of 6]). In the majority of cases, conflicts were resolved with both parents (87% [20 of 23]). In the remaining 3 cases, only 1 of the parents eventually agreed with the intended decision.
### TABLE 4  Parental involvement in decision-making

<table>
<thead>
<tr>
<th>Decision by medical team, parental permission asked</th>
<th>Decision by medical team, no parental permission asked</th>
<th>Advice of team, parents decisive voice</th>
<th>Total</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>(N=59) (49%)</td>
<td>(N=28) (24%)</td>
<td>(N=31) (27%)</td>
<td>(N=115)</td>
<td></td>
</tr>
<tr>
<td>n (%)</td>
<td>n (%)</td>
<td>n (%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

#### PATIENTS

| Diagnosis h | | | \(\chi^2 = 9.5\) |
|------------|| | |
| Congenital disorder | 34 (54) | 17 (27) | 12 (19) | 63 |
| Cancer | 15 (39) | 6 (16) | 17 (45) | 38 |
| Acute illness | 7 (50) | 5 (36) | 2 (14) | 14 |

| Age | | | |
|----|| | |
| <1 y | 22 (56) | 13 (33) | 4 (10) | 39 |
| 1-4 y | 12 (46) | 8 (31) | 6 (23) | 26 |
| 4-12 y | 13 (36) | 6 (17) | 17 (47) | 36 |
| >12 y | 9 (64) | 1 (7) | 4 (29) | 14 |

| Duration of medical care i | | | \(\chi^2 = 11.5\) |
|---------------------------|| | |
| <1 mo | 14 (52) | 8 (30) | 5 (19) | 27 |
| 1-6 mo | 24 (53) | 14 (31) | 7 (16) | 45 |
| > 6 mo | 18 (44) | 5 (12) | 18 (44) | 41 |

| Type of treatment(s) during EoLD | | | \(\chi^2 = 16.8\) (Life-Sustaining Treatment versus non-Life-sustaining treatment) |
|---------------------------------|| | |
| Cure | 21 (50) | 7 (17) | 14 (33) | 42 |
| Life-Sustaining treatment i | 47 (56) | 23 (27) | 14 (17) | 84 | <0.001 |
| Comfort care i | 10 (29) | 7 (21) | 17 (50) | 34 | 0.002 |

EoLD indicates end-of-life decision.

h In one case information on parental involvement was missing.

i \(\chi^2 = 9.5\)

j \(\chi^2 = 11.5\)

k \(\chi^2 = 16.8\) (Life-Sustaining Treatment versus non-Life-sustaining treatment)

l \(\chi^2 = 12.8\) (Comfort care only versus other types of treatment)
2.4 Discussion

Respondents of all pediatric specialties are unanimous in their preference to first discuss an end-of-life decision within their team before discussing it with the parents. End-of-life decisions are complex decisions to make. They often take place in a twilight zone in which making a sure prognosis is impossible. Extended team discussions help to reach a grounded decision by gathering and comparing different points of view. They also help emotionally by sharing opinions and emotions with colleagues. This study proves Dutch pediatric EoLDM is not a soloist activity but always a team effort.

In terms of the involvement of parents in EoLDM, our study results paint a less uniform picture. Physicians differ considerably in how they involve parents in this process. Half of the respondents choose to inform parents and subsequently ask for their permission to discontinue treatment, an approach that can best be characterized by the concept of “shared decision-making.” A quarter of the respondents choose to merely inform parents and thus protect them from feeling co-responsible, an approach referred to as “benevolent paternalism.” The remaining quarter of respondents choose to advise parents and allowed them to have the decisive voice. This approach could be characterized as “parental autonomy.” The chosen approach in EoLDM is highly influenced by 3 factors: type of decision and type and duration of treatment.

If withdrawal is considered, mainly of mechanical ventilation, physicians are less likely to leave the final decision to parents than if withholding is considered. Previous studies have shown that physicians value withdrawal as a more active and therefore more burdensome decision than withholding, although some regard them as morally equal. Physicians, feeling burdened themselves, may tend to protect parents from feeling co-responsible in these cases, which is in line with a professional attitude of benevolence. In addition, decisions to withdraw mechanical ventilation often have to be made under time pressure, because the child’s condition is rapidly deteriorating. This gives the medical team less opportunity to leave the final decision to parents. In a study by Michelson et al., parents underlined their difficulty to consider this more active decision. Only when they were convinced that their child was suffering and there was no chance of improvement, did they feel able to consider possible withdrawal of LST. Despite their difficulty, most parents did not want to be protected but preferred an approach of shared decision-making. In cases of children receiving comfort care only, physicians are most likely to present the intended decision as advice and leave the final decision to parents. In these cases, there is no chance of survival as all treatment options have failed. This unambiguous fact may make it easier for physicians to let parents be the “final arbiters” in the decision-making process, as the question no longer focuses on if but on how and when a child will die.

Our results show that most children receiving comfort care without LST are children with cancer who have been in care for >6 months. Previous studies within pediatric oncology point out that physicians and family are given the chance in this situation to build a relationship and to share information and opinions. Parents are thus better able to participate more equally in EoLDM and to even take the lead. Simultaneously, physicians are more inclined to share the overall responsibility with parents.

In the past 10 years, several studies have investigated whether parents need to be protected from co-responsibility in EoLDM. In a comparative study of PICUs in France and Canada, Carnevale et al. underline that parental guilt seems to be an individual phenomenon and has no apparent relationship with parental decisional responsibility. The conclusion of most studies is that parents should be offered the opportunity to share in the final responsibility for end-of-life decisions but without obligation.

Our study indicates that conflicts during EoLDM occur regularly, both within a team and between parents and the team. Different views on prognosis and treatment modalities are reported as the main causes of intrateam conflicts. Most parent-team conflicts arise because parents cannot (yet) agree to treatment discontinuation, either because they have a more positive view on prognosis or because of religious convictions. These findings have been confirmed in previous studies.

In our study, parents of children with congenital disorders had a more positive view on prognosis than the medical team. Hope may contribute to this more optimistic view but also the diverging opinions of parents and physicians on quality of life. As a study by Carnevale et al. underlines, the opinion of parents of disabled children and physicians differ regarding what makes the life of the child worthwhile despite its severe limitations. These differences are largely influenced by the fact that physicians mainly see the child when he or she is ill, whereas parents live with their child every day and have learned to see the possibilities in addition to the disabilities. According to the respondents, all conflicts were eventually resolved, mainly
by giving parents more time, intensifying discussions, and/or getting a second opinion. In a recent Dutch study, neonatologists reported similar conflict resolution strategies. Our study does not show whether parents truly agreed with the intended decision or merely resigned after intensive discussions.

Studies on EoLDM in Muslim communities are scarce. In a neonatal study from Oman, Muslim parents preferred to transfer all responsibility to their physician, because they did not want to be seen as having acquiesced to their child’s death. This strategy resembles the strategy chosen in our study to resolve conflicts with Muslim parents. It could be characterized as mutually agreed paternalism in contrast with benevolent paternalism, in which the transfer of responsibility is not openly discussed with parents.

Our study has several limitations. Despite our high response rate, the total number of respondents and cases was relatively low. Furthermore, our outcomes are based on the observations and opinions of the included respondents, not on direct observations of real practice. By asking detailed questions about an actual and most recent case we tried to reduce recall bias. Finally, we only recorded the physicians’ perspective. We do not know how the parents valued their involvement. Previous studies have shown that families experience less involvement and more conflicts than noticed by the medical team. Despite these limitations, our study offers new insights that have practical relevance. First, we have shown that the level of parental involvement not only varies between countries but also within a country and even within the same hospital. The approach of Dutch pediatric specialists ranges from benevolent paternalism at one end of the scale to giving parents decision-making authority and autonomy at the other. Awareness of these different approaches is important, especially if a child is transferred from one unit to another during the last phase of life or if another specialist takes over primary responsibility. In these cases, a timely and open discussion is needed between the different specialists how parents were involved and should be further involved to prevent confusion and tensions. Second, medical uncertainties are an important cause of intrateam conflicts whereas parent-team conflicts mainly arise because parents have a more positive view of prognosis or because of their religious convictions, both resulting in a wish to continue treatment. Taking more time and extending discussions help to solve conflicts within the team and with parents. Besides differences in religion between parents and team, differences in language require extra attention. Finally, we found that in almost 1 of 10 reported patients the intensification of pain and symptom management was not only intended to alleviate suffering during the dying process but also to hasten death. Regarding this aspect of EoLDM, the practice within Dutch pediatrics may actually differ from the (assumed) practices in other countries. Recent international guidelines either state that no action aimed at hastening of death is ever acceptable or that hastening of death is only acceptable as an unintended adverse effect to make the patient more comfortable. Despite these clear statements, studies by Burns et al and Inghelbrecht et al show that not all physicians and nurses find it always ethically wrong to hasten death as a goal in itself. More international research is needed before we can conclude that our finding is a unique aspect of Dutch pediatric EoLDM.

2.5 Conclusions

In a multicultural and multireligious country such as the Netherlands, decisions to withhold or withdraw treatment demand a sensitive and tailored approach by the medical team. Involving parents in end-of-life decisions according to their preferences will help them to better cope with the impending loss of their child. At the same time, the choice for a differentiated approach raises important questions, such as how can parental preferences be best explored and to what extent should parents be treated differently, with or without their knowledge? These questions require further research, directly including the parents’ perspective, as well as ongoing education and training of the medical team to anticipate on parents’ needs.

Acknowledgments

We thank all respondents for their contribution to this study. We are also grateful for the support provided by our advisory board (consisting of Bwee Tien Poli The, Hugo Heij, Frits Wijburg, Dick Tibboel, Martine de Vries, Erna Langius, Hedy Folkersma, Marc van Heerde, Mirjam Houtlosser, Christel Walhof, Petra Hollik, and Kiek Tates) and Marije Smits, Carolien van Kooten, and Hanneke Swinkels for their indispensable assistance in data acquisition and input. This study is part of a research project entitled “Tailor Made Decision Making Within Dutch Pediatrics”. It is funded by the Netherlands Organisation for Health Research and Development, which did not in any way influence the data acquisition, data analysis, or drafting of the manuscript.
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Conflicts About End-of-Life Decisions in NICUs in the Netherlands

Chapter 3

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Abstract

What’s known on this subject: Conflicts about EoL decision-making are not uncommon. Limited research in the adult and pediatric intensive care setting has shown that background and physicians’ conflict-resolving strategies vary considerably. Empirical research on decision-making conflicts in the NICU is not available.

What this study adds: Conflicts regarding neonatal EoL decisions are mainly about the child’s poor neurologic prognosis. These conflicts can virtually always be resolved by postponing the EoL decisions, thus allowing time for additional meetings, additional diagnostic tests, and second opinions.

Objective: To determine the frequency and background of conflicts about neonatal end-of-life (EoL) decisions.

Methods: We reviewed the medical files of 359 newborns who had died during 1 year in the 10 Dutch NICUs and identified 150 deaths that were preceded by an EoL decision on the basis of the child’s poor prognosis. The attending neonatologists of 147 of the 150 newborns were interviewed to obtain details about the decision-making process.

Results: EoL decisions about infants with a poor prognosis were initiated mainly by the physician, who subsequently involved the parents. Conflicts between parents and the medical team occurred in 18 of 147 cases and were mostly about the child’s poor neurologic prognosis. Conflicts within the team occurred in 6 of 147 cases and concerned the uncertainty of the prognosis. In the event of conflict, the EoL decision was postponed. Consensus was reached by calling additional meetings, performing additional diagnostic tests, or obtaining a second opinion. The chief causes of conflict encountered by the physicians were religious convictions that forbade withdrawal of life-sustaining treatment and poor communication between the parents and the team.

Conclusions: The parents were involved in all EoL decision-making processes, and consensus was ultimately reached in all cases. Conflicts within the team occurred in 4% of the cases and between the team and the parents in 12% of the cases. The conflicts were resolved by postponing the EoL decision until consensus was achieved.

3.1 Introduction

Reports describing end-of-life (EoL) practice in severely ill newborns show that death in the NICU is often preceded by the decision to withhold or withdraw life-sustaining treatment.1-4 In the Netherlands, as is the case in many other countries, the neonatologist is responsible for this decision. Usually, the parents are involved in the decision-making process: they are given detailed information, asked about their views, and are asked to ratify the proposed decision. Nevertheless, conflicts may occur between the parents and the medical team about stopping or continuing life-sup-port for severely ill newborns.2, 5 These conflicts are most likely to arise in situations where the child’s prognosis is considered to be very poor and continued treatment would involve therapies that may cause discomfort or suffering. The physicians and the parents may hold different views on which decision is in the child’s best interests. Given the irreversibility of the decision and the enormous impact on all parties involved, preventing or resolving conflicts has priority in EoL decision-making. Several theoretical discussions, guidelines, and case reports have been published on how the risk of unresolved disagreement about EoL decisions can be minimized.6-14 Empirical data about conflicts preceding EoL decisions in the NICU, however, are scarce and often lack detailed descriptions of the role of the physicians and the parents in the decision-making process.15-17 Our aim was to determine the frequency and background of conflicts between members of the medical team, and between the parents and the medical team, in EoL decisions about severely ill newborns with a poor prognosis.
3.2 Patients and methods

In the Netherlands the clinical care for severely ill newborns is centralized in 10 level III NICUs. Medical decisions are made by a multidisciplinary team led by the attending neonatologist, who is ultimately responsible. Both the parents and the physicians can initiate discussions about withholding or withdrawing treatment. EoL decisions about newborns are made irrespective of the financial status of the parents, physician, hospital, or any third party, because all costs involved in clinical and postdischarge care are covered by health insurance. The study design complied with Dutch legislation on medical privacy and medical research.

Data collection

Of the 4110 newborns admitted to Dutch NICUs between October 2005 and September 2006, we identified 359 infants who had died before the age of 2 months according to the Dutch perinatal registry. We examined the infants’ medical files and found that 340 (95%) deaths were preceded by an EoL decision and that 19 (5%) of the infants had died while receiving cardiopulmonary resuscitation. We assigned the 340 infants whose deaths were preceded by an EoL decision into 2 classifications: instable newborns who stood no chance of surviving (n = 190 [56%]) and stabilized newborns with a poor prognosis (n = 150 [44%]). This classification was based on the literature about EoL decision-making. We restricted the data collection and analysis to the stable newborns with a poor prognosis.

We extracted information from the medical files on birth weight, gestational age, day and time of death, diagnoses leading to death, and details about the decision-making process. In addition, we interviewed the attending physicians of 147 of 150 infants who died between December 2006 and October 2007. Several weeks before the interviews were to be held, we informed the physicians in the NICUs about the study and asked them if they might agree to being interviewed. We assured them that the interview would be anonymous and that all links between the physician’s name and that of the patient would be removed. In the Netherlands, previous studies on EoL practice were performed with the guarantee of legal immunity for data collection. Because this additional protection was lacking for our study, we tested the study design and feasibility in 2 pilot studies. We used a semi-structured interview to ascertain the physicians’ age, gender, number of years in the NICU, religion, and to crosscheck the data retrieved from the medical files. We asked the physicians to describe the EoL decision-making process (eg, how was the decision initiated and by whom) and to pay special attention to conflicts about EoL decisions, their source, and how they were resolved. We also asked them to mark their degree of satisfaction with the process and the extent to which the parents had influenced the EoL decision on a 5-point Likert scale (1 = strongly disagree, 5 = strongly agree). Finally, we asked each physician, “Based on your experience in the NICU, mention all the factors that you perceived as posing a risk for differences of opinion in decision-making between parents and the medical staff.” These descriptions were compressed to facilitate categorization and were read back to the physicians for approval. We requested that the physicians consulted the infants’ medical files during the interviews. All interviews were conducted by an experienced pediatrician (Dr Verhagen) and lasted between 30 and 45 minutes per patient. The interviews were recorded and analyzed separately by 2 researchers (Dr Verhagen and Ms Engels). The interval between patient care and the interviews was between 3 to 14 months.

Definitions

We defined EoL decisions as medical decisions with the effect or the probable effect that death was hastened. These included decisions to withhold or withdraw life-sustaining treatment and decisions to deliberately end the life of a newborn. Deliberate ending-of-life was defined as administering lethal drugs to end the life or to shorten the life of a physiologically stable newborn. No such case was identified in the NICUs. We defined conflict as a dispute or disagreement about an EoL decision that required some action be taken. We considered the parents to have been involved in the decision-making processes if their informed opinions about the decisions were documented in the medical files or reported in the interviews. Consensus about or agreement with an EoL decision was reached if positive confirmation of the decision was documented in the medical file or reported in the interview.
3.3 Results

In the majority of the 150 stabilized newborns with a poor prognosis, the diagnosis leading to death was asphyxia (38%), followed by congenital anomalies (17%) and sepsis or necrotizing enterocolitis (17%). The distribution of diseases leading to death was similar in all 10 NICUs. In all cases the parents had been involved in the decision-making processes. In 117 cases (78%), the physicians had initiated the processes. The physicians presented the medical teams’ decisions to the parents and asked them to ratify it. In 30 cases, equally distributed over 10 units, the medical teams solicited the parents’ views without making the teams’ recommendation explicit. In 3 cases, the parents had initiated the EoL decision-making processes. The most frequent EoL decision was to withdraw life-sustaining treatment (88%); withholding treatment occurred in 12% of the cases. Interviews were held with 80 physicians caring for 147 of the 150 newborns. Data were missing on 3 deaths, because we were unable to trace the 2 physicians involved. The number of patients cared for by 1 physician varied between 1 and 4 (mean: 1.8).

In 6 of 147 cases (4%), the decision to withdraw treatment was postponed because of conflicts within the medical teams (Table 1). In 5 of 6 cases, the source of the conflict was the child’s neurologic prognosis that was poor but not sufficiently so in the opinion of all the team members to warrant an EoL decision. In 2 cases, additional team meetings were called to achieve consensus. During these meetings the exchange of views on the future medical and neurodevelopmental sequelae resulted in consensus. In 4 cases, irreversible clinical deterioration shortly after postponing the EoL decision resulted in consensus among the team members that withdrawal of treatment was appropriate. Generally, the conflicts within the team were resolved within 34 hours (median: 24; range: 8-96 hours).

In 18 cases (12%), the EoL decision was postponed because of a conflict between the medical team and the parents (Table 2). In 14 of these cases, the medical teams suggested withdrawal of treatment mostly because of the infants’ poor neurologic prognoses, whereas the parents requested continued treatment. In 7 of the 14 cases, consensus was reached after additional meetings were called and other NICUs were asked for their opinions. The meetings were called to overcome language problems, to resolve misunderstandings on the part of the parents, or to apply family influence to help the parents decide. The second opinions were received between 2 and 5 days after they had been requested, and in all cases the results were in agreement with the first medical teams’ opinions.

In 5 of the 14 cases, the parents refused withdrawal of artificial ventilation because, in accordance with their religion, withdrawal of care was not permitted. The team members involved religious leaders and family members to help the parents with their interpretation that religion forbids withdrawal.

<table>
<thead>
<tr>
<th>No.</th>
<th>GA, wk</th>
<th>Birth weight, g</th>
<th>Cause of death</th>
<th>Age, d</th>
<th>Source of conflict in team</th>
<th>Parental opinion about treatment</th>
<th>“Intervention” leading to consensus</th>
<th>Time Interval, h</th>
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<td>820</td>
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<td>Clinical deterioration</td>
<td>24</td>
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<td>Pulmonary prognosis</td>
<td>Continuation</td>
<td>Clinical deterioration</td>
<td>52</td>
</tr>
</tbody>
</table>

Cases 1, 2, and 6 also involved conflicts between the medical team and the parents and correspond with cases 17, 3, and 9 in Table 3.

GA indicates gestational age; NEC, necrotizing enterocolitis.

<sup>a</sup> Time interval: the interval between the team’s suggested EoL decision and the final decision with parental consent.

<sup>b</sup> Extreme prematurity: <0.27 weeks’ gestational age.
<table>
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<th>Age, d</th>
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<th>Source of conflict</th>
<th>Parental opinion about treatment</th>
<th>&quot;Intervention&quot; leading to consensus</th>
<th>Time Interval, h&lt;sup&gt;a&lt;/sup&gt;</th>
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<td>Neurologic prognosis</td>
<td>Continuation</td>
<td>Clinical deterioration</td>
<td>12</td>
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Cases 3, 9, and 17 also involved conflicts within the medical team and correspond with cases 2, 6, and 1 in Table 2. GA indicates gestational age; NEC, necrotizing enterocolitis.

<sup>a</sup> Time interval: the interval between the medical team's proposed EoL decision and the final decision with parental consent.
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Cases 3, 9, and 17 also involved conflicts within the medical team and correspond with cases 2, 6, and 1 in Table 2. GA indicates gestational age; NEC, necrotizing enterocolitis.

* Time interval: the interval between the medical team’s proposed EoL decision and the final decision with parental consent.
Finally, the team adjusted their opinions and decided not to withdraw artificial ventilation but to withhold additional intensive care. They reasoned that the benefits of making EoL decisions in agreement with the parents outweighed the potential additional suffering of the infants. All 5 infants had died while connected to the ventilator. In 2 of the 14 cases, clinical deterioration of the newborns after the EoL decisions had been postponed changed the parents’ opinions.

In the remaining 4 of the 18 cases, the parents requested withdrawal of life-sustaining treatment, whereas the medical team recommended continued treatment. In 2 of these cases, additional diagnostic tests (eg, ultrasound, electroencephalogram, MRI) were ordered to increase the prognostic accuracy. The results led to consensus about withdrawing treatment. In 2 other cases, consensus was reached within 12 hours after the infants’ clinical situation deteriorated and all agreed that in the new situation withdrawal of life-sustaining therapy was appropriate.

In all 18 cases, the attending neonatologists stressed that resolving the conflicts was given priority by the members of the medical teams, because they were convinced that unresolved conflicts would hamper the parents’ grieving processes. Generally, the conflicts between the team members and the parents were resolved within 69 hours (median: 24; range: 4–336 hours).

The data on the frequency, source, and resolution of the conflicts reported in the interviews were similar to those found in the medical files. The mean score for the physicians’ overall satisfaction with the decision-making processes was 4.8 on a 5-point Likert scale. No apparent connection was found between overall satisfaction and reported conflicts. In 16 of 147 cases, physicians reported that parents had influenced the final EoL decisions substantially (score of 5 on a 5-point Likert scale). Among these 16 cases were the 5 newborns who had died while on the ventilator after the decision to withhold treatment had been made. In the remaining 11 cases, parents influenced the timing of the decision to withdraw. Withdrawal was postponed in 5 of these cases to give the parents and family members the opportunity to say their goodbyes. In the other 6 cases, the parents requested a prompt decision to end the child’s suffering. This resulted in the medical teams meeting several hours earlier than they had originally planned. The majority of physicians (63%) also indicated that they considered religious convictions that forbade withdrawal of life-sustaining treatment as an important risk factor for conflicts (Table 3).

**TABLE 3** Risk factors for conflicts between parents and the medical teams reported by 80 neonatologists in face-to-face interviews

<table>
<thead>
<tr>
<th>Frequency, n (%)&lt;sup&gt;a&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Parent-related factors</strong></td>
</tr>
<tr>
<td>• Strong religious convictions&lt;sup&gt;b&lt;/sup&gt;</td>
</tr>
<tr>
<td>• Different cultural background</td>
</tr>
<tr>
<td>• Lowest educational level</td>
</tr>
<tr>
<td>• Poor communication skills</td>
</tr>
<tr>
<td>• Language problems</td>
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<tr>
<td>• Negative previous medical experiences</td>
</tr>
<tr>
<td>• Disagreement between parents</td>
</tr>
<tr>
<td><strong>Patient-related factors</strong></td>
</tr>
<tr>
<td>• Unclear diagnosis or prognosis or both</td>
</tr>
<tr>
<td>• Absent physical signs of illness</td>
</tr>
<tr>
<td><strong>Team-related factors</strong></td>
</tr>
<tr>
<td>• Poor communication with parents</td>
</tr>
<tr>
<td>• Conflict within the medical team</td>
</tr>
<tr>
<td>• Decision-making too fast for parents</td>
</tr>
<tr>
<td>• Many different attending physicians per infant</td>
</tr>
<tr>
<td>• Little respect for parental opinion</td>
</tr>
<tr>
<td>• Strong religious convictions (of team member)</td>
</tr>
<tr>
<td><strong>Other factors</strong></td>
</tr>
<tr>
<td>• Duration of NICU admission</td>
</tr>
</tbody>
</table>

<sup>a</sup> Factors reported by >2 physicians are reported.

<sup>b</sup> Convictions that forbade withdrawal of life-sustaining treatment.
3.4 Discussion

In this retrospective, descriptive study, we investigated conflicts about EoL decisions in stabilized infants with poor prognoses in the 10 Dutch NICUs over a period of 1 year. Our study yielded 3 important findings. First, the EoL decision-making processes about infants with a poor prognosis were initiated mainly by the physicians, who subsequently arrived at the final decision together with informed and involved parents. Second, conflicts regarding EoL decisions between members of the medical team, or between the parents and the medical team, occurred in 4% and 12% of cases, respectively. The source of the conflicts frequently lay in uncertainty about the infants’ neurologic prognoses. Third, if a conflict occurred, the EoL decision was postponed until consensus was reached. The physicians’ strategies to resolve conflicts included calling additional meetings to discuss the prognoses, identifying and taking away the cause of misunderstandings on the part of the parents, and requesting second opinions.

International and national guidelines prescribe that the infant’s interests should be the main concern in neonatal EoL decision-making and that the medical team should always make decisions about withholding or withdrawing treatment in close consultation with the parents. In the Netherlands, EoL decisions are medical decisions for which doctors bear the ultimate responsibility. Our study confirmed the physicians’ prominent role in the decision-making process, and we found that the parents had always been involved. Previous studies in the Netherlands, other European countries, and the United States, reported comparable high rates of parental involvement, from 48% to 97%. Although this division of roles in EoL decision-making is considered appropriate in the Netherlands, it is contestable. The physicians’ prominent role may entail that they also control the nature and amount of information they offer the parents and that they sometimes make decisions in situations where their perspective may be limited or colored. In addition, the question remains how to define the best interests of the child, and whether the physician is the best person to make that judgment.

We found that uncertainty about the infant’s neurologic prognosis can be an important source of conflict. In the NICUs, EoL decisions were often based on a prediction of neurodevelopmental impairment. In an attempt to reliably predict impairments, all kinds of clinical scores, algorithms, and prognostic findings on ultrasound and MRI have been developed. Nevertheless, these tools have proved unable to eliminate uncertainty in individual cases. Moreover, no clear cutoff levels exist for the amount of certainty or for the level of severity of the impairment required to justify an EoL decision. Surprisingly, the majority of EoL decisions based on the infant’s poor neurologic prognosis did not lead to conflicts.

We found that in the event of conflict, EoL decisions were postponed until consensus was achieved. The first step to manage the conflict was to call additional meetings to reach consensus. Our observation that meetings were called to discuss the situation with and to inform the parents, to negotiate withholding rather than withdrawing treatment, and to use family influence is similarly described in other studies on decision-making conflicts in the context of adult and pediatric intensive care. It is interesting to note that the physicians in our study did not mention the possibility of ethics consultations or mediation to help them resolve the conflicts they encountered. Such measures have been advocated increasingly in recent literature. We were unable to determine if the consensus reached in the meetings was merely a practical compromise or if it was based on an in-depth discussion about the existing prognostic uncertainty, which was finally accepted as sufficient for an EoL decision. Studies focusing on the parental perspective of EoL decision-making indicate that parents often need more time for decisions about ending their infant’s life. These studies suggest that allowing the parents enough time is critically important to help them understand the situation and to let them assimilate with what is happening.

Our study demonstrated that in 5 cases, parents persisted in their refusal to withdraw life-support for religious reasons. The team finally decided to withhold additional treatment. This decision may have prolonged the child’s suffering. In such cases, the view held by the parents as to what is in the infant’s best interests seems to be in conflict with that held by the physician. A study from Great Britain and another from a Muslim community in Oman reported similar responses by the medical team to parental refusal to withdraw treatment. Physicians often seek agreement about EoL
decisions, because this agreement has been reported to be an important aid for the grieving process of bereaved parents. At this stage the question is raised whether acting in the child’s best interests may depend, at least partly, on the views and convictions of the parents.

In the experience of the physicians we interviewed, parental religious convictions that forbade withdrawing treatment as well as poor communication on the part of the medical team were the most important risk factors for conflicts. Awareness and understanding of the risk factors among physicians may enable them to anticipate conflicts.

We recognize several limitations to this study. The first limitation was its retrospective nature, although the fact that the medical files were available for scrutiny during the interviews may have limited potential inaccuracy in the physicians’ recall. We emphasize that the physicians’ satisfaction scores may have been influenced by the outcomes of the decision-making processes. Second, the findings were based on the physicians’ perceptions and not on those of other care providers or the parents. Third, the small number of conflicts found limited the extent to which our findings could be generalized and limited the possibilities to associate physician or patient characteristics as potential risk factors for conflicts. These concerns can probably be best addressed by combining our results with a multicenter, prospective study on conflicts in EoL decision-making for newborns. We are currently embarking on this project. A strength of our study was that all 10 NICUs in the Netherlands participated.

3.5 Conclusion

Our study showed that conflicts about EoL decisions could virtually always be resolved by postponing the EoL decisions, thus allowing time for additional meetings and diagnostic testing as well as second opinions.

Acknowledgments

This study was sponsored by the Dutch Ministry of Health, Welfare, and Sports. The funding source was not involved in the research process. We thank the neonatologists and staff of the Erasmus Medical Center (Rotterdam), Radboud University Medical Center (Nijmegen), University Medical Center (Utrecht), University Medical Center (Groningen), University Medical Center (Maastricht), VU Medical Center (Amsterdam), Academic Medical Center (Amsterdam), Isala Hospital (Zwolle), University Medical Center (Leiden), and Maxima Medical Center (Veldhoven), for providing the data for this study. We thank Dr T. Brantsma-van Wulfften Palthe in Utrecht for correcting the English manuscript.


References


Early in the evening we spoke with the doctors and asked them: what will the next step be? Deep inside we hoped that our son would decide for himself: I quit, I cannot go on living anymore. Because then you do not have to permit stopping mechanical ventilation. And you will have no doubts. As in believing that he is doing very badly, but who can be absolutely sure that his situation might not suddenly improve, unexpectedly, just after we said we would stop. During that night our son further deteriorated. The next morning a new doctor arrived. He said to us: we should not delay this any longer. At that moment we realized: no, we should not.

Quote from retrospective interview with parents
Parental perspectives on end-of-life decision-making in neonatal and pediatric intensive care patients

Chapter 4

Mirjam A. de Vos Kiek Tates Edith Leclercq Hanneke Swinkels Hugo S. Heymans Anton van Kaam Dick L. Willems Albert P. Bos

Internal publication
Abstract

Background and purpose: There is insufficient knowledge how parents experience their involvement in end-of-life decision-making and by which factors they feel guided during this process. This knowledge will add to better tailored support of parents by neonatologists and pediatric intensivists. We therefore conducted an integrative review.

Design: We searched PubMed, EMBASE, CINAHL and PsycInfo. We included qualitative and quantitative original studies that directly investigated experiences of parents of children aged 0-18 years for whom a decision to withhold or to withdraw life sustaining treatment had been made.

Main results: We identified 17 studies that met all inclusion criteria. Parents report a wide variation in how they are involved in end-of-life decision-making, ranging from having no involvement to being the sole decision maker. The majority of parents prefer to actively share in the decision to withhold or withdraw life sustaining treatment (LST). Concretely, this means that parents wish to be honestly informed, to be heard in their views and to be asked for their approval. For some parents it furthermore implies that they want to make the final decision together with their physicians. Main factors which influence parents in their decision-making are: visible suffering of their child, no chance of survival and prognosis of a very poor quality of life.

Conclusions: Most parents wish to actively share in the process of end-of-life decision-making in order to protect their child. Yet, not all parents want to make the final decision, especially not if there are uncertainties regarding prognosis.

4.1 Introduction

Children are surviving complex illnesses and surgical procedures, due to advanced technologies, continuous monitoring, and specialized training in critical care. Some critically ill children however will not survive their initial admission. A considerable number of these children will die following a decision to withhold or withdraw LST. Recent studies in European countries and in the United States have shown that in neonatal intensive care approximately 75% of deaths are now preceded by decisions to limit life support compared to approximately 50% of deaths in pediatric intensive care.\(^1\)-\(^7\) There is consensus among physicians worldwide that LST may be withheld or withdrawn if all treatment options have expired and a child has no chance of survival. Less consensus exists concerning the question whether poor quality of life can be a decisive reason to limit LST.\(^8\),\(^9\) An international comparative study showed that physicians from northern European countries largely agree with the statement that quality of life must be taken into account, whereas physicians from eastern and southern European countries tend to agree that life should be preserved at any cost.\(^10\)

Another challenging issue is the question who should decide to withhold or withdraw LST in critically ill children. In the United States and Canada parents are considered the primary decision-makers for their children, based on prevailing moral and legal traditions.\(^11\),\(^12\) By contrast, in European, Middle-Eastern and South-American countries parents are not considered qualified for making end-of-life decisions. According to the medical and public opinion in these countries, parents lack sufficient medical knowledge to make these decisions and, even more important, should be protected from the burden of responsibility.\(^6\),\(^13\)-\(^18\) Nation-specific cultural
and religious traditions are therefore important predictors of the weight given to parents in the end-of-life decision-making process (EoLDM). To date, research into EoLDM has focused on the perspectives of the medical professionals. Studies that explore parental views are scarce. Yet, in order to sensitively guide and support parents it is essential to know how parents themselves experience their involvement and how they want to be involved. The purpose of this article is to systematically trace and review original studies directly investigating parents’ perspectives. Our main questions are 1) how do parents of newborns, infants and children in different countries perceive their involvement in EoLDM, 2) how do they prefer to be involved and 3) by which factors are they guided throughout the decision-making process.

### 4.2 Methods

**Search criteria**

A search of PubMed, EMBASE, CINAHL and PsycInfo was conducted for articles published between January 2000 and July 2010, using the subject headings or text key words ‘decision making’, ‘end-of-life’, ‘withholding’, ‘withdrawing’ cross-referenced with ‘intensive care’, ‘critical care’, ‘pediatric intensive care’, ‘neonatal intensive care’, ‘PICU’ or ‘NICU’. As most end-of-life decisions are made within neonatal and pediatric intensive care, we concentrated our review on these two care settings.

**Study inclusion**

The abstracts of all potentially appropriate studies were retrieved and reviewed by four of the authors (MV, HS, AB, KT). Studies were included if they fulfilled the following criteria: (1) empirical and descriptive studies, either retrospective or prospective; (2) these studies directly investigated parental experiences and evaluations; (3) account was given of the number of participating parents; (4) participants were parents of children aged between 0 and 18 years, including neonates, for whom a decision to withhold or withdraw life sustaining treatment was considered or had been made. If studies used the same data source, only the study with the most extensive data was included.

**Data evaluation**

The final sample for this integrative review included qualitative and quantitative studies in which various methods were used, ranging from in-depth interviews to structured questionnaires. Due to this heterogeneity in primary sources, studies were coded according to two criteria: methodological rigour and data relevance. This was done on a 2-point scale (high or low) by two of the authors (MV, AB). Differences in rating were solved by consulting a third reviewer and eventually reaching consensus (KT). No studies were excluded, but studies of low rigour and/or low relevance contributed less to the analytic process.
Data extraction and synthesis

Two authors (MV, HS) independently completed a standardized data extraction form for each included full text study. Differences were resolved by discussion and, if necessary, by consulting a third and fourth reviewer (AB, KT). Extracted information included citation details, setting, number of participating parents and study design. Furthermore, it included the results of each individual study on the three themes of interest to this review: (1) parental experiences regarding their involvement in EoLDM; (2) parental preferences regarding this involvement and (3) guiding factors for parents during EoLDM. The results of the individual studies were reported in tabular form for each theme and combined as narrative. Given the heterogeneity of the included studies, meta-analysis was not attempted.

4.3 Results

The literature search identified 2729 discrete citations. After reviewing the abstracts of these papers, 37 were eligible for inclusion. The full texts of these studies were reviewed in depth. A further 20 studies were not included: 10 studies because they did not report data on our three themes of interest, 2 studies because they focused exclusively on prenatal decision-making, 2 studies because they did not directly investigate parents’ experiences and evaluations, and the last 6 studies because they made use of the same data source as included studies with more extensive data. The flow of information through this review is summarized in Figure 1.

In Table 1 we present the characteristics of the seventeen studies which were finally included. These are grouped according to type of care setting, resulting in eleven NICU-studies and six PICU-studies. Eight of the eleven NICU-studies had a qualitative design, whereas two of the six PICU-studies had a qualitative design and four a quantitative design.

Three of the seventeen included studies prospectively followed parents throughout the decision-making process. The remaining fourteen studies retrospectively investigated parents’ experiences and preferences. In the nine retrospective NICU-studies the period of time that had elapsed between the decision to withhold or withdraw LST and study participation varied considerably, from several months up to five years or more. In the five retrospective PICU-studies data were collected within a relatively short period of time after the decision to continue or discontinue LST, mostly within 0 to 18 months after the decision was considered or made.
<table>
<thead>
<tr>
<th>Source</th>
<th>Country</th>
<th>Setting</th>
<th>N parents</th>
<th>N patients</th>
<th>Study design</th>
<th>Meth rigour/ Data relevance</th>
</tr>
</thead>
<tbody>
<tr>
<td>NICU studies</td>
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<tr>
<td>Wocial, 2000 (30)</td>
<td>United States</td>
<td>1 NICU</td>
<td>20 (RR 32 %)</td>
<td>13 (all died)</td>
<td>Qualitative; retrospective; semi-structured interviews</td>
<td>High/High</td>
</tr>
<tr>
<td>McHaffie et al, 2001 (24)</td>
<td>United Kingdom</td>
<td>3 Regional Neonatal Referral Centers</td>
<td>108 (RR 67 %)</td>
<td>62 (all died)</td>
<td>Qualitative; retrospective; semi-structured interviews at 3 months and 13 months after child's death</td>
<td>High/High</td>
</tr>
<tr>
<td>Streiner et al, 2001 (29)</td>
<td>Canada</td>
<td>Follow-up of ELBW infants, treated on a NICU</td>
<td>169 versus 123 control parents (RR 86 %; 85 %)</td>
<td>169 ELBW infants versus 123 term infants (all survived)</td>
<td>Quantitative; retrospective; self-report questionnaires</td>
<td>High/Low</td>
</tr>
<tr>
<td>Brinchmann et al, 2002 (21)</td>
<td>Norway</td>
<td>NICUs, number not specified</td>
<td>35 (convenience sampling)</td>
<td>26 (10 died)</td>
<td>Qualitative; retrospective; in-depth interviews</td>
<td>High/High</td>
</tr>
<tr>
<td>Orfali, 2004 (25)</td>
<td>France and United States</td>
<td>2 NICUs</td>
<td>75 (theoretical sampling)</td>
<td>Not reported</td>
<td>Qualitative; prospective; observations and in-depth interviews</td>
<td>Low/High</td>
</tr>
<tr>
<td>Pector, 2004 (27)</td>
<td>United States</td>
<td>Support groups for parents with multiple-birth loss</td>
<td>71 (theoretical sampling)</td>
<td>71 (all died)</td>
<td>Qualitative; retrospective; semi-structured e-mail survey</td>
<td>High/Low</td>
</tr>
<tr>
<td>Ranchod et al, 2004 (28)</td>
<td>South Africa</td>
<td>Follow-up of ELBW infants, treated on 3 NICUs</td>
<td>51 (eligible parents within 2 months)</td>
<td>51 (all survived)</td>
<td>Quantitative; retrospective; structured interviews</td>
<td>High/Low</td>
</tr>
<tr>
<td>Kavanaugh et al, 2005 (23)</td>
<td>United States</td>
<td>2 hospitals providing high-risk perinatal care</td>
<td>6 (convenience sampling)</td>
<td>6 (1 died)</td>
<td>Qualitative; prospective; semi-structured interviews</td>
<td>High/High</td>
</tr>
<tr>
<td>Partridge et al, 2005 (26)</td>
<td>Australia, Hong Kong, Japan, Malaysia, Taiwan, Singapore, USA (California)</td>
<td>11 NICUs</td>
<td>327 (convenience sampling)</td>
<td>379 (16 died)</td>
<td>Quantitative; retrospective; structured interviews</td>
<td>High/High</td>
</tr>
</tbody>
</table>

NICU: Neonatal Intensive Care Unit  
PICU: Pediatric Intensive Care Unit  
RR: Response Rate  
Quantitative: Studies collecting empirical information in numerical form with measurement instruments as structured questionnaires and structured interviews  
Qualitative: Studies collecting empirical information in the form of words with instruments as unstructured or semi-structured interviews and observations  
Theoretical sampling: process of selecting parents based on emerging theoretical concepts  
Convenience sampling: process of selecting parents based on accessibility and availability  
ELBW: Extremely Low Birth Weight
<table>
<thead>
<tr>
<th>Source</th>
<th>Country</th>
<th>Setting</th>
<th>N parents</th>
<th>N patients</th>
<th>Study design</th>
<th>Meth rigour/Data relevance</th>
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<tbody>
<tr>
<td>NICU studies (continued)</td>
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<tr>
<td>Armentrout, 2009 (20)</td>
<td>United States</td>
<td>National support group of parents who experienced neonatal death of a child</td>
<td>15</td>
<td>11</td>
<td>Qualitative; retrospective; in-depth interviews</td>
<td>Low/High</td>
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<td>Einarsdottir, 2009 (22)</td>
<td>Iceland</td>
<td>1 NICU</td>
<td>53</td>
<td>29</td>
<td>Qualitative; retrospective; semi-structured interviews</td>
<td>High/High</td>
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<td>PICU studies</td>
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<td>Meert et al, 2000 (33)</td>
<td>United States</td>
<td>1 PICU</td>
<td>78</td>
<td>76</td>
<td>Quantitative; retrospective; structured telephone interviews</td>
<td>High/High</td>
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<td>Meyer et al, 2002 (34)</td>
<td>United States</td>
<td>3 PICUs</td>
<td>56</td>
<td>56</td>
<td>Quantitative; retrospective; self-report questionnaires</td>
<td>High/High</td>
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<td>Curley et al, 2003 (32)</td>
<td>United States</td>
<td>1 PICU</td>
<td>272</td>
<td>162</td>
<td>Quantitative; retrospective; self-report questionnaires</td>
<td>High/Low</td>
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<tr>
<td>Sharman et al, 2005 (36)</td>
<td>United States</td>
<td>1 PICU</td>
<td>14</td>
<td>10</td>
<td>Qualitative; prospective; semi-structured interviews</td>
<td>High/High</td>
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<tr>
<td>Carnevale et al, 2007 (31)</td>
<td>France and Canada</td>
<td>4 PICUs, 2 in France and 2 in Canada</td>
<td>31</td>
<td>19</td>
<td>Qualitative; retrospective; semi-structured interviews</td>
<td>High/High</td>
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<tr>
<td>Michelson et al, 2009 (35)</td>
<td>United States</td>
<td>2 PICUs</td>
<td>70</td>
<td>66</td>
<td>Quantitative, partly qualitative; retrospective; semi-structured interviews</td>
<td>High/High</td>
</tr>
</tbody>
</table>

NICU: Neonatal Intensive Care Unit  
PICU: Pediatric Intensive Care Unit  
RR: Response Rate  
Quantitative: Studies collecting empirical information in numerical form with measurement instruments as structured questionnaires and structured interviews  
Qualitative: Studies collecting empirical information in the form of words with instruments as unstructured or semi-structured interviews and observations  
Theoretical sampling: process of selecting parents based on emerging theoretical concepts  
Convenience sampling: process of selecting parents based on accessibility and availability  
ELBW: Extremely Low Birth Weight
Experienced involvement and the emotional consequences

Thirteen studies investigated how parents felt involved in decisions to withhold or withdraw LST for their child. Additionally, eight studies explored the correlation between involvement and subsequent emotions as regret and guilt. In Table 2 we report the outcomes of the individual studies.

While in two studies performed in the United Kingdom and in the United States the majority of parents stated that they had made the ultimate decision alone and felt capable of bearing this responsibility, in a South-African study most parents reported that their physician had made the decision, often without consulting them. In two international comparative studies - one on French and North-American NICUs, the other on French and Canadian PICUs - most French parents described a similar paternalistic approach in which their physicians decided. By contrast, most North-American and Canadian parents reported that they had made the final decision. American parents, who had to decide in an uncertain situation without sufficient emotional support by the medical team, described this as stressful and a cause of later regrets. In the second study no relationship was found between parental decisional responsibility and subsequent feelings of guilt.

In the remaining studies - performed in the United States and in six Pacific Rim countries - most parents reported that they had made end-of-life decisions together with their physicians. In three qualitative studies, all conducted in the United States, parents did not report any conflict or uncertainty when asked to share in the decision-making process on the condition that they clearly understood why withholding or withdrawing LST was considered to be inevitable and on the condition that they were convinced that they had done all they could to protect their child from suffering. According to an Icelandic study, involvement did at times contribute to feelings of guilt in parents of newborns. Yet, not taking part in the decision-making process was not considered an acceptable alternative for most parents.

Preferred involvement

Fifteen studies investigated how parents actually want to be involved in EoLDM. The results of these individual studies are also presented in Table 2.

In twelve of the fifteen studies the majority of parents preferred joint or shared decision-making over sole physician decision-making or sole parental decision-making. This preference was reported equally frequently by parents of newborns, infants and children. By sharing decisions with their physicians parents felt both in control of their child’s situation and supported in the choices that had to be made.

In eight studies, parents’ preference for shared decision-making concretely meant that they wanted to be fully informed, taken seriously in their opinions and treatment preferences and asked for their approval. Yet, these parents did not want to make the final decision and in this way bear final responsibility.

In the other four studies, parents’ preference for shared decision-making also implied that they wanted to make the ultimate decision, albeit together with their physician and thus sharing the final responsibility.

Only on NICUs in Taiwan and Singapore a substantial proportion of parents reported a preference for sole physician decision-making. According to the authors these preferences seemed highly influenced by the cultural traditions and religious beliefs in these countries. Two studies showed an interesting correlation between the medical situation and prognosis of a child and parental preferences. When the medical situation of a child was complicated or the prognosis was surrounded by uncertainty, parents wanted to share in the decision-making process but wished to leave the final decision to their physicians.

Guiding factors in parental decision-making

Sixteen of the seventeen included studies investigated by which factors parents felt guided during the process of EoLDM. The individual study results are presented in Table 3. For clarity’s sake we divided these results in three subthemes: child related - , physician related - and parent related factors.
<table>
<thead>
<tr>
<th>NICU studies</th>
<th>Experienced involvement</th>
<th>Preferred involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wocial, 2000 (30)</td>
<td>Parents felt actively involved by being well informed and having influence</td>
<td>Parents wanted control, but not all parents wanted final responsibility</td>
</tr>
<tr>
<td>McHaffie et al, 2001 (24)</td>
<td>42% perceived to bear final responsibility alone; 14% jointly with physicians</td>
<td>Most parents wanted opportunity to take responsibility for treatment limitation decisions</td>
</tr>
<tr>
<td>Streiner et al, 2001 (29)</td>
<td>–</td>
<td>Joint decision making preferred by 72%</td>
</tr>
<tr>
<td>Brinchmann et al, 2002 (21)</td>
<td>–</td>
<td>Most parents wanted active involvement but no final responsibility</td>
</tr>
<tr>
<td>Orfali, 2004 (25)</td>
<td>US parents provided with much information and final authority; French parents no authority in decision making but feeling involved due to frequent communication and strong doctor-parent relationship</td>
<td>Great variance in preferred level of involvement; Level of satisfaction not linked to level of autonomy; No parent eager to take over decision making over life or death</td>
</tr>
<tr>
<td>Pector, 2004 (27)</td>
<td>Joint decision making in most cases; Several parents reported pressure to withdraw life support</td>
<td>Physicians’ reassuring support when having to make choices as parents; Permission to choose the moment to stop treatment valued</td>
</tr>
<tr>
<td>Ranchod et al, 2004 (28)</td>
<td>41% reported physicians made decisions without consulting them; 37% after consulting them; 14% reported joint decision making; 4% made life support decisions after consulting physicians; 4% alone</td>
<td>Joint decision making preferred by 49%; Physician decision making after consulting parents in 37%; Physicians decision making without consulting parents in 8%; Sole parental decision making in 6%</td>
</tr>
<tr>
<td>Kavanaugh et al, 2005 (23)</td>
<td>Involvement merely implied giving approval; The more uncertainty, the more physicians involved parents</td>
<td>Active involvement preferred by most, but not all parents</td>
</tr>
<tr>
<td>Partridge et al, 2005 (26)</td>
<td>Majority reported collaborative decision making; Decision making by physicians alone prevailed in Australia and in Japan</td>
<td>Joint decision making strongly preferred by majority of parents; Sole physician decision making only preferred by substantial proportion of parents in Taiwan (34%) and Singapore (35%)</td>
</tr>
<tr>
<td>Armentrout, 2009 (20)</td>
<td>No regret or guilt reported over decision if convinced it had prevented further suffering</td>
<td>–</td>
</tr>
<tr>
<td>Einarsdottir, 2009 (22)</td>
<td>Involvement may lead to feelings of guilt, but not taking part is not seen as an option</td>
<td>Most parents claim a right to accept or reject life-saving treatment, because they have to live with the outcome and are the emotional experts</td>
</tr>
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(–) indicates that this study did not provide any information about this issue.
<table>
<thead>
<tr>
<th>PICU studies</th>
<th>Experienced involvement</th>
<th>Preferred involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td>Meert et al, 2000 (33)</td>
<td>In 49% of cases ultimate decision made by parents; In 15% by physicians and parents; In 29% by physicians; 71% scored their authority in making decisions as high; 71% felt physicians had right amount of authority; 22% felt physicians had too much authority and 5% too little</td>
<td>20% wanted more authority and involvement; Positive relation between parental trust in physician and satisfaction with degree of authority</td>
</tr>
<tr>
<td>Meyer et al, 2002 (34)</td>
<td>71% had little to no involvement in decision making; 55% had little to no control during the child’s final days; 76% agreed well with physicians on the decision to discontinue life support</td>
<td>24% would have made decisions differently</td>
</tr>
<tr>
<td>Curley et al, 2003 (32)</td>
<td>To not consent to ECMO not perceived as a real choice by most parents</td>
<td>72% of parents of non-survivors would consent again to ECMO versus 92% of parents of survivors</td>
</tr>
<tr>
<td>Sharman et al, 2005 (36)</td>
<td>–</td>
<td>Feelings of guilt by having to decide to withhold or withdraw life support reported by 1 parent</td>
</tr>
<tr>
<td>Carnevale et al, 2007 (31)</td>
<td>According to most French parents physician final decision maker (with parents voicing their agreement or objection); Majority of Canadian parents reported that they made the decision; No relation between decisional responsibility and guilt; No relation with outcome (survival or death of child)</td>
<td>According to most parents physicians should assume responsibility for decisions, thereby respecting parental treatment preferences; French parents more satisfied because of regular discussions with one physician and because of clear expectations regarding their (limited) role</td>
</tr>
<tr>
<td>Michelson et al, 2009 (35)</td>
<td>–</td>
<td>Parents should make decisions according to 96%; According to 50% parents should do this together with their physicians</td>
</tr>
</tbody>
</table>

(–) indicates that this study did not provide any information about this issue.
<table>
<thead>
<tr>
<th>NICU studies</th>
<th>Child related factors</th>
<th>Physician related factors</th>
<th>Parent related factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wocial, 2000 (30)</td>
<td>Observations of child's situation (more important than provided information)</td>
<td>Direct and truthful information; Test results of normal babies next to their infant's test results; Trust in physician</td>
<td>Understanding what the options meant; Acceptance that their child would probably die</td>
</tr>
<tr>
<td>McHaffie et al, 2001 (24)</td>
<td>Prospect of suffering; Visible deterioration</td>
<td>Clear, accurate and consistent information about the poor condition of the child; Concrete evidence of a bleak outcome</td>
<td>–</td>
</tr>
<tr>
<td>Streiner et al, 2001 (29)</td>
<td>If outcome uncertain, everything done until more is known</td>
<td>–</td>
<td>Religious beliefs and belief in the sanctity of life</td>
</tr>
<tr>
<td>Brinchmann et al, 2002 (21)</td>
<td>Certainty of death</td>
<td>Time for good personal communication</td>
<td>–</td>
</tr>
<tr>
<td>Orfali, 2004 (25)</td>
<td>Suffering</td>
<td>Information: in the American NICU information given in an optimistic way, in the French NICU in terms of medical certainty to protect parents from any guilt; Emotional support: in American NICU parents provided with a lot of information without emotional support, in French NICU presence of continuously available physician</td>
<td>Wish to protect child from suffering and futile care; Belief in God's will</td>
</tr>
<tr>
<td>Pector, 2004 (27)</td>
<td>Suffering</td>
<td>Careful, clear explanations and written summaries of various options' pros and cons; Facts [told] as positive as possible; Time instead of pressure by medical team</td>
<td>Not wanting child to suffer out of own selfishness; Whether other children from the pregnancy survived or not, no factor of influence</td>
</tr>
<tr>
<td>Kavanaugh et al, 2005 (23)</td>
<td>–</td>
<td>Honest and consistent information from a limited number of professionals; Written information; Support for hope, [but] no false hope; Reliance on physicians</td>
<td>–</td>
</tr>
<tr>
<td>Partridge et al, 2005 (26)</td>
<td>Pain and suffering (less reported in Japan and Hong Kong compared to other countries); Likelihood of death (less reported in Japan); Potential disability (less reported in Japan and Taiwan)</td>
<td>Physician's opinion</td>
<td>Financial issues (reported as important in Singapore and Malaysia); Religious considerations (reported as important in Singapore and Malaysia); Emotional attachment of parents to infant (particular influence not further explained)</td>
</tr>
<tr>
<td>Armentrout, 2009 (20)</td>
<td>Suffering; Certainty of death, with or without life support</td>
<td>Professionals’ support and empathy</td>
<td>Wish to protect child from prolonged suffering</td>
</tr>
<tr>
<td>Einarsdottir, 2009 (22)</td>
<td>Suffering; Inevitability of death; Living without crucial human qualities, such as any ability to take part in human communication and interaction</td>
<td>Information from medical specialists on all medical aspects, including different views and uncertainties; Individual assessment by specialists</td>
<td>–</td>
</tr>
</tbody>
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<table>
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<tr>
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<th>Physician related factors</th>
<th>Parent related factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Meert et al, 2000 (33)</td>
<td>Degree of pain and suffering; Child’s diagnosis; Expected neurological recovery</td>
<td>Physician’s information and recommendations; Time</td>
<td>Religious commitments</td>
</tr>
<tr>
<td>Meyer et al, 2002 (34)</td>
<td>Amount of pain or discomfort; Chances of getting better/likelihood to survive; Child’s quality of life; Assumed wishes of child</td>
<td>Information on overall picture; Advice and attitudes of hospital staff (not reported as very important)</td>
<td>Religious/spiritual beliefs (reported as less important than child related factors); No parents rated financial costs as important</td>
</tr>
<tr>
<td>Curley et al, 2003 (32)</td>
<td>Suffering; Inevitability of death [versus the likelihood of recovery]; Poor outcome/quality of life</td>
<td>–</td>
<td>Conviction all opportunities for survival have been tried; Wish to protect child from too much suffering; Wish not to prolong the inevitable [and thereby prolonging not only suffering of child but also own painful experience]</td>
</tr>
<tr>
<td>Sharman et al, 2005 (36)</td>
<td>Personal observations of child’s visible pain and suffering (greater weight than indirect information); [No] chance of survival; Child’s understanding and expressed wishes; Child’s perceived will to survive</td>
<td>Facts and data from various sources; Opinion and advice of physician; Time (although never enough)</td>
<td>Past experiences; Wish to protect child from suffering; Meaning parents ascribe to their disabled child’s life; Faith and belief in God’s will; Financial burden of caring for severely disabled child (n=1)</td>
</tr>
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</table>

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<thead>
<tr>
<th>PICU studies</th>
<th>Child related factors</th>
<th>Physician related factors</th>
<th>Parent related factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Carnevale et al, 2007 (31)</td>
<td>–</td>
<td>Complete explanations (without unrealistic hope); Adequate access to physicians</td>
<td>Sense of child’s condition and needs</td>
</tr>
<tr>
<td>Michelson et al, 2009 (35)</td>
<td>Suffering; No chance of getting better because of lack of effective treatments; Poor quality of life [because of] neurological dysfunction/impairment and dependence on machines</td>
<td>Physician’s prognosis; Time</td>
<td>Faith and belief in God’s will; Own intuition; Financial considerations no factor for majority of parents</td>
</tr>
</tbody>
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(–) indicates that this study did not provide any information about this issue.
In twelve studies, parents reported that their child’s visible pain and suffering was a crucial child-related factor to consider withholding or withdrawing LST.\(^{2, 1, 23, 25-28, 31, 33-37}\) The opposite also seemed to hold true. In five studies, several parents stated that in the absence of visible pain and suffering they were inclined to continue treatment even if the medical team thought it was futile.\(^{25, 31, 34, 35, 37}\)

In eleven studies parents named life expectancy as another important child-related factor. If effective treatment options were exhausted and death was inevitable, this convinced parents that treatment should not be continued. Yet, if outcomes were still uncertain, most parents preferred continuation of LST until there was more clarity.\(^{21-23, 30, 31, 33, 35-37}\)

In six studies parents reported quality of life to be a third child-related factor guiding their decision-making. If physicians predicted severe neurological impairments resulting in a very poor quality of life, this made parents consider and eventually accept the decision to discontinue LST. For most parents a very poor quality of life actually meant that their child no longer had ‘the crucial human qualities to interact and feel’.\(^{23, 27, 33-36}\)

Most parents were well aware that in their decision-making they were also influenced by physician-related factors. They felt especially guided by the quality of the information which their physicians provided. A substantial proportion of parents underlined the importance of getting clear, accurate and consistent information on the overall situation and prognosis of their child. Furthermore, they favored information provided in a positive and empathic way, but without raising unrealistic hope.\(^{23-25, 28, 31, 32, 34-37}\) Most parents highly valued the emotional support of one trusted physician.\(^{21, 22, 24, 26, 27, 31, 32, 37}\)

With regard to parent-related factors, religious beliefs were reported to be an important factor in parents’ decision-making, although of lesser importance than the child’s pain, chances of survival or quality of life.\(^{26, 30, 34-37}\) A strong belief in the sanctity of life strengthened parents in their view that all attempts should be made to save their child. At the same time, several parents indicated that their belief that God’s will prevailed also helped them to eventually accept the decision to withdraw LST. Religious beliefs seemed to play a somewhat different role in the decision-making of parents from Singapore and Malaysia. In a study which included parents living in these two countries, the authors postulated that because of their Muslim culture parents did not wish to be explicitly involved in the decision-making process because this was not considered appropriate.\(^{27}\)

Finally, four studies investigated the influence of financial considerations on parental decision-making. In studies including parents who lived in the United States, Canada, Australia and Taiwan, almost all parents reported that financial issues had not influenced their decision-making.\(^{27, 35-37}\) Only one mother stated that the financial burden of caring for a severely disabled child had influenced her decision for no full resuscitation.\(^{37}\) By contrast, a substantial proportion of participating parents from Singapore and Malaysia reported that financial costs had influenced their decision to discontinue LST.\(^{27}\)
4.4 Discussion

This review shows that there is considerable variation in how parents perceive their involvement in end-of-life decision-making (EoLDM). On ICUs studied in France, South Africa, Australia and Japan most parents reported that their physicians were the sole decision makers, whereas on the included ICUs in Hong Kong, Malaysia, Singapore, Taiwan and the west coast of the United States the majority of parents reported to have made decisions together with their physicians. Finally, on ICUs studied in the United Kingdom, Canada and the east coast of the United States most parents reported to have made the final decision themselves. None of the studies gave a conclusive explanation of these differences, but they indicate the influence of multiple factors including the cultural, religious and legal traditions in a specific country and the individual preferences of both physicians and parents. Recent studies show that physicians working within the same hospital and sometimes within the same intensive care unit involve parents quite differently in the decision-making process. It is still unclear if this diversity in involvement reflects the different personal styles of individual physicians or if it reflects the adjustment of physicians to the preferences and needs of individual parents.

Which approach do parents prefer? Our review indicates that the majority of parents of newborns, infants and children favor shared decision-making, even if this concerns the burdensome decision to withhold or withdraw life-sustaining treatment (LST). It may be postulated that parents have two important reasons for wanting to share in the decision-making process. First, in this way they can actively protect their child by making sure the process is neither going too fast (thereby denying their child the chance to show recovery) nor going too slow (thereby prolonging their child's suffering). Second, they may feel protected against later doubts and regret because they did not bear this responsibility alone.

When parents were questioned in-depth, as was done in several qualitative studies, it became clear that they attached quite different meanings to the concept of shared decision-making. For a substantial proportion of parents sharing concretely meant that they were honestly informed about their child's situation, that their views were heard and that they were asked for their approval of the proposed decision. Yet, for some parents sharing meant more than this. These parents wished to make the final decision to withhold or withdraw LST jointly with their physicians. Parents who wanted to bear this co-responsibility underlined that, although the impending death of their child caused pain and grief, these feelings did not incapacitate them to make crucial decisions in their child's best interests. The interesting correlation between uncertainties in prognosis and parents' preference to share in the process but to let their physician make the final decision to withhold or withdraw LST has also been reported in studies in adult patients. Confronted with uncertainties regarding treatment options, most adult patients wanted to participate in the decision-making process but preferred to leave the final decision to their doctor.

Only in Taiwan and Singapore a substantial proportion of the participating parents preferred to not be involved in the decision-making process at all. This seemed largely influenced by the religious and cultural traditions of both parents and physicians in these countries. Recent studies within pediatric and adult critical care arrive at similar conclusions. They indicate that on ICUs in predominantly Islamic or Catholic countries end-of-life decisions are rarely discussed with patients and their relatives because it is not considered appropriate to burden them with these decisions. Hence, patients as well as relatives do not get a chance to experience a shared decision process. In a recent Saudi Arabic study several patients nevertheless expressed a wish to participate more actively in making crucial decisions.

Regarding the question by which factors parents feel guided during the decision-making process, our review indicates that parents of newborns, infants and children have similar reasons to consider withholding or withdrawal of further treatment. Most important factors are: witnessing the child's actual pain and distress and becoming convinced that the child's quality of life will remain below a minimum threshold. For most parents this concretely implies that their child will never acquire or regain the abilities to feel and interact, not even at the most basic, non-verbal level. As is clearly outlined in two recent papers, it is often extremely difficult to predict how a child with severe impairments will develop and whether 'this future life will be so bad that it would have been better to die than to live'. The authors therefore argue that in neonates with huge uncertainties regarding prognostication parents should have the right to decide about continuation or discontinuation of treatment because they will ultimately bear the costs of that choice. This review indicates that this right may be a burden rather than a benefit. Faced with uncertainties, several parents stated that they rather wished to be protected from making the final decision and thereby from later doubts and regrets. Moreover, they needed to be repeatedly
reassured that no hasty decision would be made and that all available options had been thoroughly investigated.

This review has several limitations. The number of included studies is small and studies from Anglo-Saxon countries prevail. This limits the generalizability of the outcomes to other countries. Furthermore, not all studies clearly defined the concepts used, especially the concept of ‘shared decision-making’. This makes it harder to verify whether parents applied the same meaning to the questions in interviews and questionnaires as the researchers. Finally, there was considerable variation between studies in time between the decision-making period and data collection. We do not know how this affected parents’ evaluation. We are well aware that because of these limitations the outcomes that we describe should be interpreted with caution and require further comparative research among larger groups of parents of different patient groups. Inclusion of more countries, especially from Asia, Africa and South-America, will provide a better insight into the influence of cultural, religious and socio-economic factors on parental EoLDM. Our review indicates that in poorer countries parents’ decisions to withhold or withdraw LST are influenced by lack of insurance and other financial resources. In a time in which the gap widens between rich and poor countries, this tendency needs to be closely monitored in the best interests of children and parents.48

4.5 Conclusions

Parents participating in the included studies report a broad variation in how they are actually involved in EoDM. At the same time, they largely agree in their preference to actively share in the decision-making process. This does not automatically imply that they also want to make the final decision to withhold or withdraw LST. The more uncertainties regarding prognosis, the more parents indicate that they want to be protected from bearing this responsibility. Parents’ strongest motive to share in the decision-making process is their wish to protect their child from further suffering in his or her last phase of life. Parents highly value frequent discussions with a regular physician during the decision-making process. These dialogues make parents feel involved, also if they do not (wish to) bear any decisional authority.24-26, 29, 31, 34 Furthermore, the resulting trust in their physician helps parents to accept the unacceptable: that further treatment is futile and that their child will most probably die.

Acknowledgments

We are grateful for the indispensable help of Arnold Leenders and the valuable advice of Frans Plötz, Martin Offringa, Hans Reinders and Petra Hollak.
References


The shared perspective

Physician: ‘Your child is what matters most to us. As doctors we have to take care that what parents wish for is something their child can still endure.’

Mother (crying): ‘It is a real dilemma. I do not want my son to suffer, not for one percent. And if everything has been tried already, I do not want things to be continued. But I want to have him near me for some more time. Yet, I know there is no time anymore.’

Physician: ‘We do not need to stop now. You need time to say goodbye. We’ve still got time.’

Quote from meeting between a mother and the physician in charge
Chapter 5

Talking with Parents about End-of-Life Decisions for their Children

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Albert P. Bos
Frans B. Plötz
Marc van Heerde
Bert M. de Graaff
Kiek Tates
Dick L. Willems

Accepted for publication by Pediatrics
Abstract

What’s known on this subject: Retrospective studies have shown that the majority of parents, independent of their country of origin, prefer a shared approach over a paternalistic approach or an informed approach when an end-of-life decision has to be made for their child.

What this study adds: In actual conversations parents act in line with their preference for a shared approach. This contrasts with the ‘some sharing’ approach of physicians who carefully prepare parents for an end-of-life decision already being made by the medical team.

Background and objective: Retrospective studies show that most parents prefer to share in decisions to forgo life-sustaining treatment (LST) from their children. We do not yet know how physicians and parents communicate about these decisions and to what extent parents share in the decision-making process.

Methods: We conducted a prospective exploratory study in two Dutch University Medical Centers.

Results: Overall, 27 physicians participated and 37 parents of 19 children for whom a decision to withhold or withdraw LST was being considered. Forty-seven conversations were audio-recorded, ranging from 1 to 8 meetings per patient. By means of a coding instrument we quantitatively and qualitatively analyzed physicians’ and parents’ communicative behaviors. On average, physicians spoke 67% of the time, parents 30% and nurses 3%. All physicians focused primarily on providing medical information, explaining their preferred course of action and informing parents about the decision being reached within the team. Only in 2 cases parents were asked to share in the decision-making. Despite their intense emotions most parents took considerable effort to actively participate in the conversation. They did this by asking for clarifications, offering their preferences and reacting to the decision being proposed (mostly by expressing their assent). In the few cases where parents strongly preferred LST to be continued, the physicians either gave parents more time or revised the decision.

Conclusions: We conclude that parents are able to handle a more active role than they are currently being given. Parents’ greatest concern is that their child might suffer.

5.1 Introduction

After the death of a child, parents’ lives are never the same again. Despite their immense grief, most parents find a way to continue living as best as they can. The factors that support them in coping with the loss of their child have received growing attention.2-15 Recent studies show that most parents find it helpful to share in their child’s daily care during the dying process. Most of them also find it helpful to share in decisions to withhold or withdraw life-sustaining treatment (LST), decisions which often precede a child’s death.2, 3, 5, 9, 11, 16-20 Parents’ perception of a shared approach may even lower their grief in the long term.21 The extent to which parents want to share in the decision-making process differs from parent to parent. Some want to share in all stages of the decision-making process, including exchanging information, deliberating about preferred options, and making the final decision together (Table 1). Others want to share information and preferences with their child’s physician but feel it as too great a burden to make the final decision. Therefore, recent studies have concluded that the way parents are involved should be highly tailored to their individual preferences and needs.2, 16, 22-31 What we do not yet know is how parents are actually being involved in end-of-life decision-making. Little information is available about how these decisions are made in PICUs. We therefore performed a prospective exploratory study aimed at answering two questions: How do physicians and parents communicate about decisions to withhold or withdraw LST, and to what extent do parents share in the decision-making process?
5.2 Methods

Study setting and inclusion of parents

We conducted our study on the PICUs of 2 Dutch university medical centers. Both PICUs are combined medical and surgical tertiary facilities, including all specialty care except that involving prematurely born infants. The institutional review boards gave their approval for the study. To identify eligible conversations, the main researcher (M.A.V.) contacted the supervising pediatric intensivist twice a week. Upon being informed about an intended conversation in which a decision to withhold or withdraw LST would be discussed with parents, she contacted the physician who would be speaking with the parents. This physician then informed the parents about the study and asked for their consent. Parents were informed about the main purpose of the study: to help physicians to improve their communication with parents about treatment choices in critically ill children. It was stressed that parents had the full liberty to refuse and could end their study participation at any time. We strived to achieve a large variation of cases according to the following patient-related characteristics: age, gender, prognosis, pre-existing neurological damage, total duration of care, length of PICU stay. Similarly, we sought variation according to the following parent-related characteristics: gender, ethnic background, first language, religion, marital status, and length of physician-family relationship. The first inclusion period lasted from April 2008 until April 2009, the second from April 2010 until April 2011 (see Table 2 for details, including the consent rates of parents and physicians).

Coding and analysis

The recorded conversations were transcribed verbatim and anonymized. Transcripts were then uploaded to MAXQDA (VERBI GmbH, Berlin, Germany). We performed a search in PubMed for schemes used to codify physicians’ and parents’ communication in end-of-life decisions. In the pediatric literature we did not find any such schemes. In the adult medical literature our search eventually led to 2 relevant and useful publications. First, Charles et al described a conceptual framework for decision-making in the medical encounter, consisting of different analytic stages: information exchange, deliberation, and deciding on treatment to implement. Second, White et al described a coding instrument to assess the extent of shared decision-making in relation to specific physician communication behaviors.
In the first draft of our coding scheme we incorporated the 3 analytical stages and the coded physician behaviors. After having closely read the first 10 transcripts, we elaborated the coding elements describing physician behaviors and added coding elements describing parents’ communication behaviors. After additional refinement of this extended coding scheme, transcripts were coded by 2 researchers (M.A.V. and B.M.G). This included a recoding of the first 10 transcripts. Conflicting views about the applied codes were resolved by discussion. Next, we composed worksheets containing all retrieved conversation segments per applied code over all cases. We qualitatively and quantitatively analyzed this material and thoroughly discussed the results in several discussion rounds with all researchers. Coding scheme and worksheets are available upon request.

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<thead>
<tr>
<th>TABLE 2</th>
<th>Quantitative context of the study</th>
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<tbody>
<tr>
<td></td>
<td>PICU 1, n (%)</td>
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<tr>
<td><strong>First inclusion period April 1, 2008 - April 1, 2009</strong></td>
<td></td>
</tr>
<tr>
<td>• Included cases</td>
<td>5</td>
</tr>
<tr>
<td>• Missed cases&lt;sup&gt;b&lt;/sup&gt;</td>
<td>3</td>
</tr>
<tr>
<td>• Deaths (percentage of patients admitted to both PICUs)</td>
<td>9</td>
</tr>
<tr>
<td>• Patients dying after a decision to forgo LST (percentage of total number of deaths)</td>
<td>6 (67%)</td>
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<tr>
<td><strong>Consent rate of parents</strong></td>
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<tr>
<td>• Consent rate of physicians</td>
<td>100%</td>
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<tr>
<td>• Consent rate of nurses&lt;sup&gt;d&lt;/sup&gt;</td>
<td>100%</td>
</tr>
<tr>
<td><strong>Second inclusion period April 1, 2010 - April 1, 2011</strong></td>
<td></td>
</tr>
<tr>
<td>• Included cases&lt;sup&gt;e&lt;/sup&gt;</td>
<td>0</td>
</tr>
<tr>
<td>• Deaths (percentage of patients admitted to both PICUs)</td>
<td>12</td>
</tr>
<tr>
<td>• Patients dying after a decision to forgo LST (percentage of total number of deaths)</td>
<td>8 (67%)</td>
</tr>
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<sup>a</sup> 13 of 15 children died within 12 mo after the decision to forgo LST had been made, and 2 of 15 children survived.

<sup>b</sup> In these cases, conversations with parents had taken place before the researchers were informed.

<sup>c</sup> Analysis of the 15 cases included in the first period showed that cases with the following characteristics were underrepresented: pre-existing neurological damage in the child, long-term physician-family relationship or religious parents. In the second period we included 4 more cases with ≥ 1 of these characteristics by purposeful sampling.

<sup>d</sup> 2 of 4 children died within 12 mo after the decision to forgo LST had been made, and 2 of 4 children survived.

<sup>e</sup> Of the 39 parents we approached, 2 parents (1 couple) refused to participate.

<sup>f</sup> Of the 28 physicians we approached at the start of our study, 1 physician refused to participate.

<sup>g</sup> A total of 26 nurses participated. They were present at 32 of 47 meetings.
5.3 Results

Characteristics of participants and of the decision-making process

Overall, 37 parents of 19 patients and 27 physicians participated. Table 3 lists their demographic characteristics. In Supplemental Table 8 we present the main characteristics of the decision-making process per patient.

Speaking time and contribution of nurses

Forty-seven conversations were audio-recorded. The number of meetings ranged from 1 to 8 meetings per patient, with a median of 2 meetings. The mean duration of meetings was 30 minutes. On average, physicians spoke 67% of the time, parents 30% and nurses 3%. Whenever a nurse was present, his or her contribution to the conversation always occurred at the end of the meeting and consisted of a combination of emotional and practical support, as indicated by remarks such as: “This must be difficult to hear” or “We’ll arrange that you can both stay with your child tonight”. The nurse then accompanied parents back to their child’s room or got parents coffee and stayed in the conference room with them for some time.

Communication behaviors throughout the decision-making process

In all but 1 case, physicians and parents went through all stages of the decision-making process: providing and receiving information, deliberating about whether to withhold or withdraw LST, and reaching a final decision. In none of the cases this was a linear chronological process. Instead, within a conversation, physicians and parents moved back and forth between the stages of information exchange and deliberating before either reaching a decision to withhold or withdraw LST at the end of the meeting or postponing this decision to a subsequent meeting. The next meeting typically start with an extensive update about the child’s situation and a renewed deliberation about the pros and cons of treatment continuation or discontinuation. In 6 cases it took 1 meeting to reach a final decision, in 7 cases 2 meetings and in the 5 most complex cases ≥3 meetings. In the remaining case the child died just after the first meeting. In this meeting the physician cautiously announced that there could come a time when continuation of LST had to be considered futile.

Stage I. Providing and receiving information

Table 4 gives an overview of the specific communication behaviors physicians and parents showed in the first stage of decision-making, the number of cases in which we identified these behaviors, and illustrative quotes extracted from the transcripts.

Although physicians occasionally started the meeting by explaining its goal and by presenting a short agenda, in most meetings he (or she) immediately started with an extensive and detailed update about the child’s current serious condition, the grave prognosis, and the remaining treatment options. Much of this information was repeated several times throughout the meeting. Most physicians emphasized that everything was being tried to overcome the patient’s life-threatening problems, but that treatment was becoming increasingly burdensome for the child.

In one third of the cases, the physicians asked parents to provide their perspectives about their child’s situation, especially their observations about symptoms of discomfort and their child’s ability to make any form of contact. None of the parents were invited to share their views about their child’s prognosis or the remaining treatment options.

In half of the cases, parents themselves took the initiative to ask clarifying questions throughout the meeting. These questions showed that they wanted to understand exactly what was happening to their child. Furthermore, they specifically asked which treatments were being given to their child, whether they were working, and whether everything was being done to protect their child from pain and other symptoms. Toward the end of the meeting most physicians did ask the parents if they had any additional questions. Occasionally physicians asked them to summarize their understanding of the situation. In these cases, the parents typically demonstrated good understanding of the key issues.
<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Patients (N=19, n (%))</th>
<th>Parents (N=37, n (%))</th>
<th>Physicians (N=27, n (%))</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Age (y)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0-1</td>
<td>5 (26)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1-4</td>
<td>8 (42)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>4-12</td>
<td>4 (21)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>12-17</td>
<td>2 (11)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Mean age (y)</td>
<td>4</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td><strong>Gender</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>4 (21)</td>
<td>19 (51)</td>
<td>12 (44)</td>
</tr>
<tr>
<td>Male</td>
<td>15 (79)</td>
<td>18 (49)</td>
<td>15 (56)</td>
</tr>
<tr>
<td><strong>Main diagnosis</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Congenital disorder</td>
<td>11 (58)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Acute illness</td>
<td>2 (10)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Neurotrauma</td>
<td>2 (10)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>SIDS</td>
<td>2 (10)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Cancer</td>
<td>1 (6)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Perinatal asphyxia</td>
<td>1 (6)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td><strong>Total duration of medical care</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>0 – 24 h</td>
<td>1 (6)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1 – 7 d</td>
<td>4 (21)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1 – 4 wk</td>
<td>4 (21)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1 – 6 mo</td>
<td>2 (10)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>&gt; 6 mo</td>
<td>8 (42)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td><strong>Long-term physician-family relationship</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>3 (16)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>No</td>
<td>16 (84)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td><strong>Interval between final decision and death</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 24 h</td>
<td>5 (26)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1-7 d</td>
<td>8 (42)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>1-4 wk</td>
<td>0 (0)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>&gt; 1 mo</td>
<td>3 (16)</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Still alive</td>
<td>3 (16)</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

* Defined as: there was 1 physician in charge who had treated the patient for > 6 mo and had had regular conversations with the parents about the patient’s diagnosis, prognosis and treatment plan over this period (with a minimum of 3 times a year).

b This relatively large number is explained by 2 factors. First, on both PICUs intensivists mainly chaired the meetings with parents, either accompanied by another intensivist or by a specialist also involved in the child’s care. Second, successive meetings were frequently chaired by a different intensivist.

(−) indicates not applicable.
TABLE 4  Stage I: Providing and receiving information

<table>
<thead>
<tr>
<th>Coded behaviors</th>
<th>From physicians to parents</th>
<th>From parents to physicians</th>
<th>From physicians to parents</th>
<th>From parents to physicians</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Cases (N=19)</td>
<td>n (%)</td>
<td>n (%)</td>
<td>Cases (N=19)</td>
</tr>
<tr>
<td>1. Asking for information about actual situation and treatment effects</td>
<td>6 (32%)</td>
<td>“Perhaps you can tell me more about what has actually happened. Because as far as I know, up till this morning your daughter was completely healthy?”</td>
<td>8 (42%)</td>
<td>“The EEG showed no activity, but we were told that he shows some activity when you look at him. What causes this activity? Where are these signals being made? In his cerebral cortex?”</td>
</tr>
<tr>
<td>2. Providing information about actual situation and treatment effects</td>
<td>18 (95%)</td>
<td>“Her lunges have much difficulty to get enough oxygen and to transport this to all her organs. Slowly we are approaching the last remaining options. And we have started with the last option today by giving her extra medication.”</td>
<td>11 (58%)</td>
<td>“I observe my son every day and I see that he is frightened. And when he gets his feeding by his PEG he becomes nauseated.”</td>
</tr>
<tr>
<td>3. Asking for information about prognosis</td>
<td>0 (0%)</td>
<td>–</td>
<td>10 (53%)</td>
<td>“She is in a coma now. But what is the chance that she will recover from it and how damaged will she be?”</td>
</tr>
<tr>
<td>4. Providing information about prognosis</td>
<td>18 (95%)</td>
<td>“All his organs will suffer. But especially the organs which need most energy - his brain, lunges and heart – will be the first to collapse. And we have no cure.”</td>
<td>1 (5%)</td>
<td>“I don’t expect him to show a miracle anymore. Everything has been tried.”</td>
</tr>
<tr>
<td>5. Asking for information about remaining options, including pros and cons</td>
<td>0 (0%)</td>
<td>–</td>
<td>11 (58%)</td>
<td>“Are there no antibiotics anymore you can give him? Because he has so much sputum.”</td>
</tr>
<tr>
<td>6. Providing information about remaining options, including pros and cons</td>
<td>19 (100%)</td>
<td>“If your son is able to breathe well, we will try to get him out of the ICU and home as soon as possible. If it does not work out and breathing will prove difficult, it is not right to intubate him again, because he has an underlying illness which cannot be cured.”</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
<tr>
<td>7. Asking for summary</td>
<td>3 (16%)</td>
<td>“I would like to begin by asking you to tell me what you remember from our conversation earlier this afternoon.”</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
<tr>
<td>8. Providing summary</td>
<td>0 (0%)</td>
<td>–</td>
<td>3 (16%)</td>
<td>“In short: we heard that our son is brain damaged and that none of his functions can be saved anymore.”</td>
</tr>
<tr>
<td>9. Inviting to ask additional questions</td>
<td>15 (79%)</td>
<td>“Have you understood everything? Do you have any questions? Please, let us know.”</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
</tbody>
</table>

(–) indicates not applicable.
Stage II. Deliberating

In Table 5 we present the specific communication behaviors physicians and parents showed in the second stage of decision-making, the number of cases in which we identified these behaviors, and illustrative quotes.

In none of the cases did the physicians ask the parents how they preferred to be involved in the decision-making process. In 1 case, the parents spontaneously expressed their preference and explained that they wanted to fully share in the decision-making process, including making decisions about whether to discontinue LST together with their physician.

Almost all physicians at some point explained which decision they would prefer. They presented these preferences as team preferences rather than personal preferences. In most cases, physicians stated that according to the team’s shared opinion it would be best if LST was withheld or withdrawn, whether in the short term or longer term. Depending on the child’s specific condition and prognosis, physicians explained this preference as follows: Despite full support it was becoming clear that the child was deteriorating very fast and would die soon (in 7 of 19 cases); continued use of LST could not overcome the underlying problems and was only causing the child suffering (in 6 of 19 cases); or re-instituting LST would put the child’s already fragile quality of life under even more pressure (in 4 of 19 cases). In the remaining 2 cases, the physicians explained in the first meetings that they preferred LST to be continued. In their opinion more time should be taken to await the child’s possible recovery. This preference contrasted with the parents’ preference to withdraw LST to spare their already severely disabled child more suffering. Over time the condition of the children deteriorated and in later meetings the physicians underlined that LST had clearly become inappropriate and therefore should be withdrawn.

In the majority of cases (in 16 of 19 cases) parents presented their preferences about additional treatment, often without being invited to do so. One third of these parents (5 of 16 cases) expressed a preference for treatment to be continued or restarted, in contrast to the preference presented by the physician to withhold or withdraw LST in the near future. In these cases, the parents explained that they were not yet certain that all had been tried and that they still thought and hoped that their child could recover. Several parents added that their religious beliefs strengthened this hope. Two thirds of the parents (11 of 16 cases) emphasized that they did not prefer treatment to be continued at all cost. In most cases this preference was in line with that of the physician, in 2 cases (already mentioned above) the parents’ preference that LST would be withdrawn was not yet shared by the physicians. The parents of both children underlined that their wish to try everything to save their child by now was outweighed by their wish to protect their child from pointless suffering and from living a life they considered inhumane.

All parents reacted emotionally when the physicians indicated that a decision to withhold or withdraw LST could become inevitable or had become inevitable. These emotions varied between anxiety, dismay, grief, confusion, and disbelief. The parents of the 2 children mentioned above described how they felt a strange combination of deep sadness and relief for their child’s sake. Several other parents described how they felt trapped in the dilemma of wanting their child to be near them as long as possible and wanting to spare their child more suffering. Parents’ often intense emotions did not prevent them from taking the initiative to ask clarifying questions and to present their preferences. While most physicians acknowledged the parents’ emotions and were empathic, none of them explored these feelings further. Occasionally, physicians addressed parents’ needs for emotional and social support.

Stage III. Reaching a decision

Table 6 gives an overview of the specific communication behaviors physicians and parents showed in this third stage, the number of cases in which we identified these behaviors, and illustrative quotes.

In 2 cases the physicians asked the parents which decision they thought should be made. In these conversations, a differentiated “package of decisions” was formulated, including withholding of future resuscitation, withholding of LST in case of severe, lasting problems, and initiating LST in case of “bridgeable” problems such as an epileptic seizure or a pneumonia. In the remaining cases, the physicians did not invite parents to share in making the final decision. Instead, during the (last) meeting physicians explained that the medical team had reached the conclusion that LST had become “futile” and should therefore be forgone. In almost half of these cases, physicians added that this decision was a medical decision and not a decision parents should make.
### TABLE 5  Stage II: Deliberating

<table>
<thead>
<tr>
<th>Coded behaviors</th>
<th>From physicians to parents</th>
<th>From parents to physicians</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Cases (N=19)</td>
<td>Illustrative quotes</td>
</tr>
<tr>
<td></td>
<td>n (%)</td>
<td></td>
</tr>
<tr>
<td>1. Asking which role in decision making would be preferred</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
<tr>
<td>2. Providing information which role in decision making would be preferred</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
<tr>
<td>3. Asking about values and preferences regarding continuation or discontinuation</td>
<td>4 (21%)</td>
<td>“If he were to catch a severe pneumonia, what would you say? Should we continue to treat him in the ICU?”</td>
</tr>
<tr>
<td>4. Providing information about values and preferences regarding continuation or discontinuation</td>
<td>17 (89%)</td>
<td>“Technically we can do almost everything to keep your son alive. But in fact we would only be prolonging his suffering and not giving him a real future.”</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
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<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Expressing objections to the other’s preferences</td>
<td>3 (16%)</td>
<td>“Of course I understand that this is against your parental intuition. Parents never want treatment to be stopped for their child. But everything we do should be in the interests of your daughter, not in our own interests.”</td>
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<tr>
<td></td>
<td></td>
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<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Inviting to share emotions</td>
<td>2 (11%)</td>
<td>“You were upset by what I was telling you, weren’t you? Please tell me.”</td>
</tr>
<tr>
<td>7. Expressing emotions of grief, fear, despair, and frustration</td>
<td>0 (0%)</td>
<td>–</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
TABLE 5  Stage II: Deliberating

<table>
<thead>
<tr>
<th>Coded behaviors</th>
<th>From physicians to parents</th>
<th>From parents to physicians</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Cases (N=19)</td>
<td>Cases (N=19)</td>
</tr>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>8. Acknowledging emotions</td>
<td>15 (79%)</td>
<td>1 (5%)</td>
</tr>
<tr>
<td></td>
<td>“It is totally understandable that this frightens you.”</td>
<td>“It is difficult, isn’t it? It is also hard for his nurses.”</td>
</tr>
</tbody>
</table>

(-) indicates not applicable.

In the majority of cases parents’ reactions made it clear that they did not feel taken by surprise and understood the inevitable need to withhold or withdraw LST. Nevertheless, in 4 cases the parents repeated their strong wish that LST be continued. After acknowledging how difficult this process was for the parents, all physicians suggested a follow-up meeting for additional discussion. In this meeting the physicians again emphasized the child’s critical condition and the duty of all caregivers to not prolong the child’s suffering. Yet, they also gave the parents the opportunity to clarify their points of view. The parents of 2 children eventually agreed that there remained no other choice than to withdraw LST (in both children death was clearly imminent). In the other 2 children, the physicians and the parents agreed that not all life-sustaining treatments would be withheld.

At the end of the (last) meeting, all parents expressed their great concern that their child might suffer in the process of dying. In turn, the physicians promised that everything would be done to assure the child’s comfort and peace. Moreover, several parents asked whether it would be possible to let their child regain consciousness to speak with him or her for the last time.

### Extent of shared decision-making

In Table 7 we provide an estimate of the extent of sharing which we identified in our study. In most cases the interaction between the physicians and the parents could best be described as “some sharing”. This implied that information and preferences were shared, mostly at the parents’ initiative, but the decision-making was not.

#### 5.4 Discussion

When an end-of-life decision has to be made for a child, most parents - independent of their country of origin - seem to prefer a shared approach by their physicians over a paternalistic approach or an informed approach. Our findings indicate that in actual conversations parents act in line with this preference. Most parents in our study made an effort to actively participate in the decision-making process, especially when their initial preferences regarding the course of treatment did not correspond with those of the medical team. By contract, most physicians were primarily focused on carefully preparing the parents for the decision to withhold or withdraw LST. This approach seemed motivated by their dual wish to not overburden parents with too much responsibility while letting them grow towards accepting the inevitability of this decision.

Studies investigating end-of-life communication with families in adult ICUs have reported a comparable focus by physicians on providing medical technical information. What our study adds is the insight that parents appreciated receiving this detailed information, as evidenced by their reactions and their questions in return. It may be postulated that the provision of relevant information helps parents to grasp what is happening to their child and regain some feeling of control. Although in most cases there seemed to be the right balance between parents’ and physicians’ communication behaviors in the first decision-making stage (exchanging information), our data also suggest that physicians’ and parents’ behaviors in the second stage (deliberating) and third stage (reaching a final decision) were often out of balance. In these cases a better balance could have been reached if the physicians had actively invited the parents to explain their points of view. Moreover, although the physicians firmly believed that they had the final responsibility for whatever decision was reached, the parents would have appreciated and preferred an affirmation that the decisions were being reached in collaboration with them and with respect for their views and values.

Parents’ striving to fully share in all decision-making stages as well as physicians’ striving to act as the final decision-maker both seemed strongly motivated by an urge to protect. Parents wanted to guard their child’s life and, even more important, to protect their child from suffering. Physicians wished to guard the child’s life and well-being, but also wished to protect the parents from (later) worries and guilt.
TABLE 6  Stage III: Reaching a decision

<table>
<thead>
<tr>
<th>Coded behaviors</th>
<th>From physicians to parents</th>
<th>From parents to physicians</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Cases (N=18)</td>
<td>Illustrative quotes n (%)</td>
</tr>
<tr>
<td>1. Asking whether and which decision should be made</td>
<td>2 (11%)</td>
<td>“We could reach a point where it is necessary to perform cardiac massage or start mechanical ventilation. Should we do this or shouldn’t we?”</td>
</tr>
<tr>
<td>2. Informing about decision being reached</td>
<td>16 (89%)</td>
<td>“We think his condition is so severe that we have reached a point in time where continuing of ventilation is not appropriate anymore. This is the only treatment still keeping him alive. We want to stop the ventilation. This means that he will die.”</td>
</tr>
<tr>
<td></td>
<td>8 (44%)</td>
<td>“Of course we will discuss the decision [to withdraw ventilation] with you, but we never let parents decide. That would be impossible for you in a situation like this.”</td>
</tr>
<tr>
<td>4. Asking for agreement regarding proposed decision</td>
<td>5 (28%)</td>
<td>“We are not going to resuscitate him for a second time because that would be too much damage for his already damaged brain. Do you agree with this decision?”</td>
</tr>
<tr>
<td>5. Expressing agreement or assent</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>6. Expressing dissent</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>7. Expressing worries about suffering</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

In the remaining 19th case the child died before this last stage was being reached in the conversations with the parents. (–) indicates not applicable.
TABLE 7 Extent of sharing between physicians and parents

<table>
<thead>
<tr>
<th>Process of decision-making</th>
<th>Characteristics of a shared process</th>
<th>Extent of sharing identified in our study</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stage I: Providing and receiving information</td>
<td>1. Exchanging information:</td>
<td>+++ (at the parent’s own initiative)</td>
</tr>
<tr>
<td></td>
<td>1.1. Physician informs parents about actual situation, prognosis, treatment options and their risks and benefits</td>
<td>+++ (in most cases not elicited by physicians, but at parents’ own initiative)</td>
</tr>
<tr>
<td></td>
<td>1.2. Parents inform physician about their observations and considerations</td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>2. Helping parents understand:</td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>2.1. By inviting to ask questions</td>
<td>+++</td>
</tr>
<tr>
<td></td>
<td>2.2. By checking understanding</td>
<td>++</td>
</tr>
<tr>
<td>Stage II: Deliberating</td>
<td>3. Discussing which role parents prefer to have in the decision-making</td>
<td>at the parent’s own initiative</td>
</tr>
<tr>
<td></td>
<td>4. Discussing treatment preferences:</td>
<td></td>
</tr>
<tr>
<td></td>
<td>4.1. Physician expresses preference</td>
<td>+++</td>
</tr>
<tr>
<td></td>
<td>4.2. Parents express preference</td>
<td>+++ (in most cases not elicited by physicians)</td>
</tr>
<tr>
<td></td>
<td>4.3. Exchanging underlying values and deliberations</td>
<td>++ (especially when parents have different preferences than physicians)</td>
</tr>
<tr>
<td>Stage III: Reaching a decision</td>
<td>5. Making the final decision together</td>
<td>+</td>
</tr>
<tr>
<td></td>
<td>6. Reaching agreement (eventually) about the most appropriate decision</td>
<td>+++</td>
</tr>
<tr>
<td>Overall</td>
<td></td>
<td>++ / +++</td>
</tr>
</tbody>
</table>

Yet, this protection was not in line with what parents asked for, as indicated by their efforts to actively participate in all decision-making stages.

In this respect our findings have important practical implications. First, they may help physicians to become more alert to parents’ communication behaviors and to how these behaviors interact with their own. This alertness will help them to tailor their approach to parents’ approaches in the different decision-making stages. Second, our findings may also prove helpful in the ongoing debate, at least in the Netherlands, regarding the role of minors and their parents in end-of-life decision-making. According to Dutch law, the judgment about the appropriateness of treatment continuation or discontinuation is a medical professional judgment in which the views of minors and their parents should be taken into account.\(^{45}\) It could seriously be questioned whether this law is still suited to the actual practice in which parents want to share in deciding whether treatment has become inappropriate.

In our study, most parents eventually expressed their assent to the decision being presented to them, convinced that there remained no other choice. In a minority of the cases the parents kept their resistance, mainly because they still hoped and believed that their child’s situation would improve. In these cases the physicians did not put pressure on the parents. Instead they either gave them more time to be with their child and to witness the ongoing deterioration despite full support or revised their decision to meet the parents’ wishes. This outcome confirms the finding of earlier studies that physicians make considerable efforts to prevent potential conflicts from escalating.\(^{46-48}\) Seen in this light, the choice of physicians to come forward to the wishes of the parents who disagreed could be characterized as a conflict-solving strategy. At the same time it could be considered unfair that parents who express their strong objections may influence the decision-making process to a greater degree than parents who comply with the decision being proposed. We do not yet know which emotional effects compliance or noncompliance may have for parents in the long term. On one hand, it can be hypothesized that parents who comply to the recommendation to forgo LST may later feel that they have given up too soon and blame themselves for not expressing their doubts and concerns. On the other hand, parents who resist the recommendation to forgo LST may later feel guilty about not allowing their child to die peacefully. Follow-up studies of these long term effects are needed to better support individual parents throughout the decision-making process and in their later bereavement after their child’s death.
Our study suggests that in most cases parents’ intense emotions of anxiety, grief, and distress did not hinder them from asking relevant questions and from clearly explaining their considerations and preferences. It could even be argued that these emotions, especially the parents’ fear of losing their child and their anxiety of seeing their child suffer, heightened their alertness and awareness. In this respect there was no apparent difference between parents whose child had acutely fallen ill compared to parents whose child had a congenital disorder. This finding implies that the capacity of parents to understand complex issues should not be underestimated, despite their stress and grief. At the same time, it remains important that physicians use clear, unambiguous words, do not lose themselves in too many details and complex hypothetical scenarios, and keep encouraging parents to ask questions and present their points of view.40, 43, 49-53

Our study has several limitations. An important limitation is that our study was restricted to two medical centres, which was unavoidable given the logistical demands of the study. Moreover, we cannot rule out that the presence of a researcher influenced the course of the conversations. The greatest strength of our study is that we were able to explore in detail how physicians and parents communicate about end-of-life decisions. In order to do this we developed a coding instrument to systematically explore their communication behaviors. This instrument needs greater validation, but may prove useful in international comparative research of actual end-of-life discussions by parents, physicians, and nurses. Although our study did not find evidence that nurses contributed to the decision-making during the “key conversations” we observed, this does not imply that nurses did not play an important role in other settings. Sometimes it was the nurses who first raised the issue of redirecting care in multidisciplinary team meetings. In addition, at the bedside nurses often spoke with parents about the information they had received earlier that day and explored parents’ reactions. It may be that the views that parents expressed in follow-up meetings were views that they had come to develop in these conversations at their child’s bedside. In future research it would be of interest to observe both formal and informal discussions between parents and caregivers and compare their impact on the decision-making process.

5.5 Conclusions

Most parents in our study took considerable effort to actively participate in the decision-making process, especially when their initial preferences did not correspond with those of the medical team. By contrast, most physicians were primarily focused on carefully informing the parents and not on actively inviting them to share in the decision-making. In most cases parents’ intense emotions of anxiety, grief, and distress did not hinder them from asking relevant questions and in explaining their considerations and preferences. Physicians made considerable efforts to prevent potential conflicts from escalating. Our study reveals that parent feel the same double duty as physicians, making certain that everything is tried to save their child’s life while on the other protecting their child from suffering, without sufficient quality of life. What parents fear most is that their child will suffer in the process of dying.

Acknowledgments

We are very grateful for the trust of the participating parents, physicians and nurses. Moreover, we thank Hanneke Swinkels for her valuable assistance in the data collection phase of this study and Prof. Hugo Heymans, Henk Galekamp, Prof. Hans van Goudoever, Petra Hollak, Prof. Johan Legemaate en Prof. Ellen Smets for their helpful comments and advice. This study is part of a research project entitled ‘Tailor-made decision-making within Dutch paediatrics. How parents can be better involved in end-of-life decisions for their children’. This project is funded by the Netherlands Organisation for Health Research and Development (ZonMw). ZonMw did not in any way influence the data acquisition, data analysis, or drafting of this manuscript.
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<th>Patient</th>
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<th>Neurological impairment</th>
<th>Total duration of medical care (length of last stay in PICU)</th>
<th>Number of audio-recorded meetings</th>
<th>Number of physicians involved in meetings</th>
<th>Conflicting views with parents</th>
<th>Final decision</th>
<th>Outcome</th>
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SIDS, sudden infant death syndrome

* Withdrawing LST implied withdrawing mechanical ventilation in all cases.

* Withholding LST implied the withholding of resuscitation and of mechanical ventilation in all cases (unless stated differently).
Parents who wish no further treatment for their child

Chapter 6

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Antje A Seeber
Sjef K M Gevers
Albert P Bos
Ferry Gevers
Dick L Willems

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Abstract

Background: In the ethical and clinical literature, cases of parents who want treatment for their child to be withdrawn against the views of the medical team have not received much attention. Yet, resolution of such conflicts demands much effort of both the medical team and parents.

Objective: To discuss who can best protect a child’s interests, which often becomes a central issue, putting considerable pressure on mutual trust and partnership.

Methods: We describe the case of a 3-year-old boy with acquired brain damage due to autoimmune-mediated encephalitis whose parents wanted to stop treatment. By comparing this case with relevant literature, we systematically explore the pros and cons of sharing end-of-life decisions with parents in cases where treatment is considered futile by parents and not (yet) by physicians.

Conclusions: Sharing end-of-life decisions with parents is a more important duty for physicians than protecting parents from guilt or doubt. Moreover, a request from parents on behalf of their child to discontinue treatment is, and should be, hard to over-rule in cases with significant prognostic uncertainty and/or in cases with divergent opinions within the medical team.

6.1 Introduction

In children with severe acquired brain damage, the decision if and when treatment may be foregone is never simple. Because scientific research is scarce in this patient group, knowledge about the correlation between type and degree of damage, on the one hand, and duration and level of recovery, on the other, is still lacking. Accordingly, physicians’ opinions about the period of time during which substantial neurological recovery may occur and treatment should be continued can be quite diverse. Physicians’ views also vary on what may be considered a substantial level of recovery and an acceptable quality of life. An uncertain prognosis complicates end-of-life decision-making for medical teams and also for parents. It confronts physicians with the question as to what extent parents should be involved in the decision-making process. Guidelines published by national paediatric societies give contradictory advice about how to best involve parents. Some recommend that all decision-making authority should be referred to parents as the child’s main representatives, thereby favouring an informed approach, others recommend that physicians should retain the final authority, thereby favouring a paternalistic approach. A recent guideline, published by the Dutch Paediatric Society, recommends a semi-shared approach in which parents should be actively involved in the decision-making process while the physician in charge is responsible for making the final decision.

Ultimately, the discussion how best to involve parents comes down to one central dilemma: how can physicians best fulfil the ‘double ethical duty’ of sharing end-of-life decisions with parents while protecting them from too much responsibility and later regrets. This balancing act becomes even more delicate when physicians and parents disagree. And while most literature centres on the situation in which parents demand (allegedly) futile care for...
their child,\textsuperscript{14–17} the opposite situation, though by no means uncommon, is much less discussed.

In this paper we first describe the decision-making process for a 3-year-old boy with severe acquired brain damage where the parents wanted to stop further treatment contrary to the views of the medical team. Next, we compare this case with recent literature. Finally, we formulate answers to the following questions: (1) what are the pros and cons of sharing end-of-life decisions with parents in cases where treatment is considered futile by parents and not (yet) by physicians; (2) how should the double ethical duty of physicians, as previously described, be fulfilled in such cases and (3) what can be learnt from our analysis for the guidance of parents in end-of-life decision-making? We collected data through chart review, through audio recordings of team discussions and of meetings between parents and the medical team, and through in-depth interviews with parents and primary care professionals within 6 months after the patient’s death. The institutional review board of the Academic Medical Centre of Amsterdam reviewed our study and gave their approval. After having read the first draft of this paper, the parents and primary care professionals gave their consent for publication. It was the parents’ explicit wish to use their son’s true name.

6.2 Case Report

Shortly after his third birthday Bruce became seriously ill with symptoms of high fever and vomiting. Two days later he had his first epileptic seizure, from which he did not regain full consciousness. He was acutely admitted to the local hospital. On suspicion of herpes simplex encephalitis (HSE), the medical staff started treatment with intravenous aciclovir. Because of his multiple drug-resistant seizures Bruce had to be transferred to the paediatric intensive care unit (PICU) of the affiliated university medical centre the next day. Here treatment with aciclovir and antiepileptic drugs was continued. MRI showed extensive areas of abnormal intensity in the right and left temporofrontal lobe, consistent with HSE. After 14 days of treatment Bruce could still not speak clearly and had diminished strength in his left limbs. However, he was seizure-free and fully alert again. In discussion with Bruce’s parents the team decided to transfer him back to the local hospital. A few days later his parents were allowed to take him home. Over the week his condition improved further. Yet at the beginning of the following week there was a sudden deterioration.

Father: “Back home, he started talking again, he rode his bike, he played and he walked. Then, one day, we could tell by the look in his eyes that things were wrong and we knew we were heading for trouble again.”

Mother: “Yes, you knew what was happening.”

Quote from retrospective interview with parents

That same day Bruce was admitted to the local hospital again. There his neurological functioning rapidly deteriorated: he lost his ability to talk and walk and had swallowing difficulties. Again, he had seizures and involuntary limb movements. Most of the time, he was unable to recognise his surroundings. Extended diagnostic investigation did not show the cause of this second deterioration. Bruce’s condition became unmanageable when he began to show signs of uncontrolled behaviour like spitting and biting his parents. He was referred back to the PICU for further treatment.
Father: “In the local hospital Bruce had to be physically restrained or else we had to be there 24 hours a day to take care of him. You don’t even chain your dog that way! So at that point we said: “If the neurologist does not expect any further improvement, we don’t want him to have heart massage or ventilation anymore. Nothing at all.” We signed a contract in which this was stated. But after his transfer to the academic hospital this contract had to be annulled. There they wanted to intubate him again and try something new. It was our last and only hope, so we felt we had to grasp it.”

Quote from retrospective interview

As soon as Bruce was back in the academic hospital, the frequency of seizures rapidly increased as did his involuntary movements. There were more and more indications that Bruce had autoimmune-mediated encephalitis, triggered by the herpes virus. This kind of autoimmune reaction is rare, especially in children, and evidence-based treatment guidelines are therefore not available. After an extensive team discussion and consultation with experts abroad, it was decided to start treatment with an immunosuppressant agent (azathioprine) and plasmapheresis under ventilation and sedation.

Physician: “We discussed our options. And we decided to start plasmapheresis in combination with several other drugs. This approach was based on theoretical arguments and a case report of a patient whose neurological condition had improved substantially with this therapy. I don’t think these were strong arguments, but we all felt the need to do something in light of what was happening.”

Quote from retrospective interview with one of the physicians involved

Bruce’s parents hesitantly went along with this treatment plan. Their worst fear was that it would prolong their son’s suffering after all that he already had had to endure. Moreover, they were worried that their son would not recover to an acceptable degree.

Mother: “How much damage do you see in his brains? Which damage is permanent? How will he be?”

Physician: “That is a very difficult question and I don’t have the answer yet. The MRI shows damage, but we can’t tell whether this damage is permanent and what will be the consequences. But we think that we should try to fight this infection and his reaction with all possible means. Unfortunately we cannot say if and how he will recover.”

Father: “I understand that, but do you feel that it has any purpose at all? What’s been happening over the past two weeks is a mystery to us. Is it not too late?”

Quote from meeting between parents and one of the physicians involved

After having consulted more experts, the medical team decided to apply the proposed treatment for 2 weeks and then evaluate its effects and discuss further steps. This time schedule was explained to the parents in the course of several meetings. Yet, none of the physicians could state explicitly what would be evaluated and what further steps would possibly be taken.

Mother: “Each time we had a different physician and they all explained the situation somewhat differently. It was already complicated, but this made it even more incomprehensible. At a certain moment we bought an audio-recorder so that we could listen again to what had been said.”

Quote from retrospective interview

After 2 weeks, there seemed to be no positive effects, although Bruce’s condition was hard to evaluate. Reducing his sedation immediately intensified his epileptic seizures, unrest and discomfort. In a team discussion, several members raised their doubts about whether treatment burdens by now might outweigh the uncertain benefits. Other members underlined that it was still too soon to forego treatment. In their opinion more time was needed to await a possible recovery.

Physician 1: “With regard to his future we are still only talking about his basic functions, like swallowing and keeping his airway open. And we also don’t know if we can stop his epileptic seizures.”

Nurse: “And what about the consequences of all the medication that he gets.”

Physician 2: “We need to consider his quality of life as well.”
Physician 1: “I agree. But we should make decisions at the right moment. We really should take sufficient time. We owe it to this little boy.”

Quote from team discussion

In this intensive discussion the staff members eventually agreed to prolong the installed treatment for at least another week. Bruce’s parents felt overwhelmed by this sudden prolongation.

Mother: “We had understood that he would be extubated today. We don’t see any improvement now and we didn’t last Tuesday. So we thought: today it will all be over. But now we hear that his treatment will be continued. [...] It becomes harder and harder to retain a little hope. It is like a roller coaster.”

Physician: “All these emotions are very tiresome. But try to remember that we will look at Bruce every day to check his responses. He may become more restless again. I know: it is hard to see any positive signs right now. [...] And even if his situation improves, it will be a road with many obstacles.”

Mother: “This powerlessness doesn’t make it any easier.”

Quote from a follow-up meeting between parents and a different physician

The parents’ exhaustion intensified their feelings of confusion, anger and grief. They shared these feelings with different members of the nursing staff. Some members had the impression that the parents’ despair was caused by the prospect of having to take care of a son who would not recognise them anymore, who would be unable to make any contact and who could not be fed because of extreme unrest and epileptic seizures. To others it seemed as if the parents were already in mourning and had given up hope that their son would survive.

Physician: “Perhaps the time is coming near to discuss whether we still consider this treatment appropriate. But not at this moment, while we are still fighting for his life.”

Nurse: “We need to involve the parents in this fight. Yet, it almost seems as if they have already given up. They have repeatedly asked me whether it is justified what we are doing. They are far ahead of us.

Physician: “I don’t think ‘ahead of us’ is the right way to say it.”

Nurse: “Perhaps not, but the parents are in the process…”

Physician: “... of saying goodbye. Still, I think it is our duty to treat this situation with the utmost care. You can only make this decision once.”

Quote from a successive team discussion

A second brain scan (MRI) after 3 weeks of treatment showed extended irreparable loss of brain tissue in the temporofrontal lobes. In a multidisciplinary meeting with all specialists involved it was decided to gradually decrease the sedatives and subsequently extubate Bruce. It was also decided not to reintubate him again if he was unable to breathe without support. The parents fully agreed with this plan and again emphasised their wish that everything should be done to spare their son further suffering. Early that same afternoon Bruce was extubated. Shortly thereafter his epileptic seizures increased and he began to show signs of serious discomfort. At the parents’ urgent request the dose of morphine was increased and propofol was started.

Mother: “Bruce clearly became troubled.”

Father: “I urged repeatedly to give him more morphine. But they answered that he was already getting so much.”

Mother: “We had a new team of doctors who did not yet know Bruce. It seemed as if they wished to give him a last chance. Yet it was long over.”

Quote from retrospective interview

Eventually Bruce became respiratory insufficient. Late that evening he died on his mother’s lap. That same night the parents took his body home.
6.4 Discussion

According to Dutch law, parental permission is required before starting (any) new treatment in patients aged between 0 and 12 years of age, except in acute, life-threatening situations. Physicians are considered to be responsible for the decision that treatment has become futile and should therefore be stopped or not started anew. For this decision no parental permission is required. Yet it is recommended that parents are asked if they agree with this decision.18 Recent studies have shown that end-of-life decisions regularly give rise to conflicts between medical staff and parents.14-17 In most cases, parents want treatment to be continued, whereas physicians consider this futile. The bottom line in such cases is that physicians are not obliged to perform any treatment they consider inappropriate according to professional standards. The reverse situations where parents demand discontinuation of treatment while physicians think it too early are rarer. Such cases are also less often discussed in the ethical literature.19 Yet the question whether physicians have the right to continue treatment against the express wishes of parents is important and has direct clinical relevance. It also provokes the ensuing question whether it is psychologically and morally acceptable to place the burden of the decision to discontinue treatment on the parents. Below we describe the main arguments to protect parents from difficult treatment decisions, on the one hand, and to allow them to play a substantial, even decisive role in this process on the other. Additionally, we apply these arguments to our case.

Is parental involvement in the interest of the child?

It has often been argued that parents should not have a substantial say in end-of-life decision-making because, as laymen, they lack sufficient medical knowledge to contribute equally to this difficult process. This might especially be true in complex cases with an uncertain neurological prognosis as in the case of Bruce.20-23 Furthermore, it is argued that parents in addition to lacking medical expertise, also lack power because of their subordinate position. Physicians will thus always succeed in getting parental permission, even if this demands ‘subtle negotiations or simply presenting only one alternative’;22 In addition, in children with acquired brain damage the acute character of the illness will initially overwhelm parents and hinder their ability to understand the complex information.24-25 This combination of reasons, it is argued, makes parents less able than professionals to define and guard the child’s best interests. In the retrospective interviews about our case, professionals mentioned another reason why active involvement of parents in the decision-making process would not be in the interest of the child. Respondents stated that the decision to withhold or withdraw treatment has to be a rational decision, based on facts. Yet in their opinion the parents’ preferences were largely influenced by their emotions of grief, despair and exhaustion, which did not add to, but merely hindered, objective decision-making. This line of reasoning can be criticised. Bruce’s parents were able to substantiate their opinion with realistic observations of the facts—namely, Bruce’s lack of interaction, the continuing seizures and his discomfort as soon as sedation was reduced. These observations were very much in line with those witnessed by the medical staff. Furthermore, would medical teams be able to make a purely objective and rational decision on continuation or discontinuation of treatment if parents were not involved? In recent years several studies have compared end-of-life decision-making by physicians from different centres and countries. These studies show how end-of-life decision-making is always influenced by the physicians’ personal values. These values are coloured by religion, culture, previous experiences and emotions.11, 22, 26-29 Objective, ‘value-free’ decision-making is thus an illusion.

In conclusion, we cannot be sure that the involvement of parents is in the best interests of the child, but the same applies to healthcare providers. Physicians, together with parents and other professionals involved, can only try to define the child’s best interests to the best of their ability at each stage of the child’s treatment and guard and redefine these interests whenever necessary.30

Is parental involvement in the interest of the parents?

The psychological state of parents may be considered as an argument against active parental involvement from yet another perspective. Asking parents for their permission to withhold or withdraw treatment may cause them to feel responsible for their child’s death. This may give rise to feelings of guilt afterwards.2, 31 But guilt may also arise when parents feel that they have become responsible for a life that has become extremely burdensome for their child and lacks any comfort, interaction or pleasure, as could have been the case for Bruce. Several studies have retrospectively explored how parents themselves evaluate their role in end-of-life decision-making. In the majority of
these studies, both parents of younger and older children evaluated active involvement as helping. Most of the parents who had not been actively involved stated that they would have preferred a greater share in the decision-making process in order to better protect their child’s well-being. In retrospect, Bruce’s parents still regretted not having been given the opportunity to share in making the central decisions. They felt that it would have enabled them to better protect their son from the suffering he had to endure, especially his severe discomfort. In previous studies parents underlined the importance of receiving honest and consistent information, also about remaining uncertainties. Furthermore, they felt it was helpful when this information was accompanied by their physician’s personal arguments for and against possible course(s) of action.

In summary, the available evidence does not seem to point to an overburdening of parents when allowing them to share in the decision-making process. Even if there were evidence to suggest serious feelings of guilt in parents, this does not, from an ethical point of view, imply that physicians should protect parents by assuming all responsibility themselves. The death of a child is a terrible tragedy and parental feelings of guilt may be an unavoidable part of that tragedy. We have to conclude that diminishing the involvement of parents, with a view to protecting them, is hardly defensible.

**Conflicting interests**

Authors like Beauchamp and Childress have stipulated what ‘good parents’, when trying to decide in the best interests of their child, should do—namely, ‘weigh the benefits and burdens of a possible course of action, considering such factors as relief of suffering, preservation or restoration of function, and quality and extent of the life sustained’. Like parents, healthcare professionals have a moral duty to act in their patients’ best interests and on the basis of their patients’ preferences, even if these conflict with their personal views. As a result, parents and professionals are obliged to monitor each other’s decisions to ensure that these are in the interest of the child. This monitoring process may have two obvious contrasting consequences. On the one hand, if physicians make decisions which disregard the child’s well-being in the short and long term, parents will want to disqualify those decisions. The repeated requests of Bruce’s parents not to prolong his treatment, based on their observations of his suffering and the continuing deterioration in his quality of life, can be seen as such an attempt. On the other hand, parents may be disqualified as proxy decision-maker if they make a choice which clearly threatens their child’s best interests.

In a recent publication, Diekema goes one step further and argues that parental decision-making authority may only be limited if parents place their child at significant risk of serious preventable harm. This may occur if parents request futile treatment which would prolong their child’s suffering or if they refuse treatment which would clearly benefit their child. We do not think that in our case—characterised by prognostic uncertainty, a lack of evidence-based treatment options and by divergent opinions within the medical team—the parents could or should have been limited in their role of proxy decision maker. Their wish to discontinue life-sustaining treatment was meant to prevent further harm to their son and in light of all uncertainties it cannot be defended that this wish increased the risk of preventable harm. This implies that the parents’ views and preferences should not have been overruled, as happened in this case.

It remains a point of discussion whether the interests of the child can and should be separated from the interests of his or her parents. This issue is reflected in expert opinions which underline that parents, not physicians, will have to live with the consequences of the decision to continue or discontinue treatment. If treatment is continued, parents will have to care for a child who might have very severe impairments. If treatment is discontinued and the child dies, they will carry that loss for the rest of their lives. These experts therefore are of the opinion that parents should be the ‘final arbiters’ in end-of-life decision-making. In our case some team members felt that the most important reason why Bruce’s parents wanted life-sustaining treatment to be stopped was to protect themselves from the further sorrow of seeing their child deeply damaged. In the retrospective interview the parents indicated they had been concerned about their ability to protect their son 24 hours a day from hurting himself because of his uncontrolled behaviour. It would have meant that he had to continue living a life of constant suffering, a suffering against which they had wanted to protect him all along.

A study by Sharman et al shows that most parents are well aware of the internal struggle between their need to avoid their own agony and suffering and their wish to do what is best for their child. An open and sensitive discussion of this struggle will be helpful, to support parents, and also to encourage a more transparent decision-making process.
6.6 Conclusions

In this paper we have tried to answer three questions concerning the case of a child whose parents wanted discontinuation of his treatment against the views of the medical team: (1) What are the pros and cons of sharing end-of-life decisions with parents and do the pros outweigh the cons? (2) How can the double ethical duty of involving parents and protecting them at the same time be fulfilled in practice? (3) What can be learnt from our analysis of this case for the guidance of parents in end-of-life decision-making?

Do the pros outweigh the cons?

Upon reviewing our discussion of the pros and cons, we conclude that the arguments in favour of involving parents in end-of-life decision-making outweigh the arguments against. Making decisions in the best interests of their child will always be hard for parents because of the emotional state they are in. Nevertheless, most parents can cope with an active role and find it helpful. There is growing evidence that it was not the parents who actively participated but the parents who felt excluded and therefore unable to sufficiently protect their child who were prone to later feelings of doubt and guilt. This is clearly illustrated by our case. For most parents active involvement means that their opinion is integrated into the decision-making process and their agreement is sought. For some parents it also implies making the final decision, either alone or together with their physicians.22, 23, 32, 33, 44-46

In cases characterised by significant uncertainties about effective treatment options and prognosis - and therefore without clear criteria by which physicians can justify one specific course of action47 - we argue that parents should have the opportunity to make decisions in close cooperation with their child’s physician. Some parents will prefer treatment to be continued in order to have (more) certainty that every chance has been taken. Others will prefer treatment to be discontinued at an earlier stage than members of the medical team to spare their child the undue burden of treatment which in their opinion has become inappropriate.

How can the double ethical duty of sharing and protecting be fulfilled?

We have stated that by giving parents the opportunity to share in the decision-making process they may be protected from later doubts and regrets. However, there will always be parents who wish to be protected from having to share in the decision-making process. Physicians should of course respect this wish. At the same time parents’ preferences about their role may change over time. Well-considered involvement of parents therefore also implies that physicians regularly check whether parents feel their points of view are respected and that they are involved to the right extent.5, 32, 35, 36, 46

In this paper we have explored yet another double ethical duty which physicians face: the duty to share decisions with parents versus the duty to protect their patient’s best interests. Parents may and should be overruled when they refuse treatment that is clearly beneficial for their child. Parents may and should also be overruled when they request strictly futile treatment that will prolong their child’s suffering. Both situations occur rarely. Most conflicts between parents and physicians arise when there is no unequivocal best course of action, situations as in the case of Bruce, characterised by prognostic uncertainties and, often, substantial variation in physicians’ attitudes towards treatment continuation or discontinuation.48

In this ‘grey zone’ of decision-making we think there is no ethical justification for over-ruling parents’ preferences. Instead we conclude that the physician’s duty to protect the child’s best interests implies his or her duty to actively share this process with parents and - if parents wish so - to let them make the final decision.49

What can be learnt from our analysis for the guidance of parents?

1. Let parents participate in the process of end-of-life decision-making by honestly discussing the remaining treatment options, including their pros and cons, and inviting them to express their views and preferences.
2. Check regularly whether parents feel involved to the right extent.
3. Give parents the opportunity to decide whether treatment should or should not be stopped in cases characterised by prognostic uncertainties and/or divergent opinions within the medical team.
4. Ensure a consistent process and do not suddenly reverse a decision or change the agreed time path. Assign one doctor who supports parents through the entire process.
5. Trust parents in being the true proxies of their child. Parents may only be disqualified as proxy decision-makers if they indisputably threaten their child’s best interests.

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This last chapter is partly based on the closing lecture which I held for an audience of physicians, nurses and other professionals working in the Emma Children’s Hospital AMC on October 3rd 2014.

The following link gives access to this web lecture which is subtitled in English: http://webcolleges.uva.nl/Mediasite/Play/2afbb4fa227e4f6a9107a74718e53d951d.
Introduction

Decisions to forgo treatment are among the most difficult decisions to make, from a medical, ethical, legal and psychological point of view. They confront us with the limits of our medical knowledge, with moral dilemmas without best answers, and with feelings of powerlessness, injustice and grief. Making end-of-life decisions for a child is an even more burdensome process. There is a substantial difference between deciding to forgo treatment together with an adult patient who can look back on a life worth living or making this same decision with parents for a child whose life has only just begun. The most difficult part of making this decision is that it concerns a life and death decision for another human being. Moreover, in many cases we will never know for 100% sure that this decision was the best decision to make for this particular child and that this moment was the best moment to do so.

This research project began with exploratory interviews with parents and physicians facing these decisions. Their perspectives differed, self-evidently. The personal relationship between parents and their child is of a different order than the professional relationship between a physician and his or her patient and the fear of losing your child is a much deeper and more devastating fear than the fear of losing your patient. But despite these different perspectives, parents and physicians expressed a similar need: how can we best reach a shared decision and what can we learn in that respect from the experiences of other parents and physicians. These questions became the starting point for our research project which officially started in 2008 and lasted until 2015. Throughout this period we performed four successive studies: an interview-study among Dutch neonatologists; a survey among Dutch pediatric intensivists, oncologists, neurologists,
neurosurgeons and metabolic pediatricians by means of a questionnaire; an integrative review of international empirical studies among parents; and an observational study of end-of-life conversations between parents and physicians in two University Medical Centers. Looking back on this project, what did we find? In the next paragraph I will recapitulate the main research questions and present our overall answers. I will then evaluate this project in light of the literature and critically review our methodology. I will end this chapter with a compact overview of recommendations to improve the process of end-of-life decision-making in children and to better involve parents in this process. To this overview I will add recommendations for further research.

Integrating the outcomes

This project aimed at answering eight key questions. We investigated these questions in the four successive study phases by means of different methods and by first focusing on the physicians’ perspective, then on the parents’ perspective and, finally, on their combined perspectives. This closing chapter provides the opportunity to integrate the individual study outcomes and present overall conclusions. Below, I will present these conclusions in a question and answer format. At the end of each section, I will refer to the specific chapters on which this conclusion is based. Some conclusions are based on our review. If this is the case, references to these studies are added.

1. **How and why do Dutch physicians decide to withhold or withdraw treatment in a child?**

All pediatric specialists participating in the interview study and in the survey reported that they extensively discuss a decision to withhold or withdraw treatment within their own team. Main aim of this exchange is reaching inter-collegial consensus. If all team members or the vast majority consider treatment to have become ‘medisch zinloos’, i.e. futile, this conclusion is then discussed with the child’s parents.

According to the observational study, in conversations with parents physicians concretize the reasons why one or all types of treatment have become futile in one of the following ways:

1. Despite full support the child is deteriorating very fast and will die soon;
2. Continuing life-sustaining treatment will not help anymore to overcome the underlying problems. Instead of benefiting, it will burden the child and prolong his or her suffering;
3. Re-starting life-sustaining treatment will put the child’s already fragile quality of life (because of multiple severe impairments) under even more pressure and will cause suffering.

Physicians’ message that treatment has become futile is always followed by what it implies, namely that this treatment will have to be forgone in the short term. The concrete arguments being presented by physicians can be divided into two main categories: reasons based on ‘quantity of life’ (1), meaning that treatment might sustain life but not for long because the child will almost certainly die, and reasons based on ‘quality of life’ (2 and 3), meaning that there are serious concerns about the child’s suffering, now and
in future, and/or about the child’s survival in a severely impaired state. In the observational study, the following treatments were eventually withheld or withdrawn: cardiopulmonary resuscitation, mechanical ventilation and (seldom) antibiotics, chemotherapy and extracorporeal membrane oxygenation. In none of the cases, withholding or withdrawing artificial feeding was taken into consideration. The cases presented in the survey as well as the cases observed in the observational study illustrate that it often takes several meetings with parents before a decision is being reached. Moreover, in many cases successive decisions are being made. Especially in children with congenital disorders, the decision to withhold life-sustaining treatment (based on quality of life arguments) is often followed by the decision to withdraw all treatment (based on the fact that death seems imminent). Occasionally a child survives, against all expectations. The decision to withhold life-sustaining treatments is then reinstalled. This decision is again based on quality of life arguments. (Chapters 2, 3 and 6)

2. How do physicians involve parents in decisions to withhold or withdraw treatment?

According to our survey, Dutch physicians involve parents very differently in end-of-life decisions for their children. This ranges from an approach in which the physicians do all of the decision-making, to an approach in which parents make the final decision, based on the information the physicians have provided to them. Between these two extremes lies a shared approach in which physicians and parents exchange their information and preferences to eventually reach a joint decision. We found that physicians may choose very different approaches despite the fact that they work within the same medical center and sometimes within the same medical team. Statistical analyses showed that the approach physicians chose in a most recent case was not significantly influenced by personal characteristics like their gender, years of experience or religious affiliation. Instead, we did find a significant correlation between the chosen approach and the type of decision being considered in this case, the type of treatment the child received and the length of the relationship between the physician and the child and his or her parents. This implied that physicians facing a decision to withhold life-sustaining treatment (based on quality of life arguments) tended to protect the child received and the length of the relationship between the physician and the child and his or her parents. This implied that physicians facing a decision to withhold life-sustaining treatment (mainly mechanical ventilation) tended to protect the child and his or her parents. This implied that physicians facing a decision to withhold life-sustaining treatment (based on quality of life arguments) tends to a shared approach in which they decided together with the parents which life-sustaining treatments should be withheld or withdrawn. This often concerned children with congenital disorders. Critically ill children are often treated by more than one pediatric specialist. If these specialists vary in the way they let parents participate in the decision-making process, this may cause confusion and frustration in parents. (Chapters 2 and 5)

3. What are the main reasons for parents to consider withholding or withdrawing their child’s treatment?

In the observational study and in the studies included in the review, parents mentioned three key reasons why they either had decided or would decide to withhold or withdraw treatment from their child. First, witnessing their child’s suffering, as measured by his or her visible pain and other symptoms like shortness of breath, extreme motoric unrest, discomfort and anxiety. Second, knowing that their child’s death had become inevitable and imminent. And finally, their conviction that their child’s quality of life would remain below the minimum threshold. In that respect parents mentioned two crucial criteria: the estimation that their child would never be able (anymore) to express basic emotions and the estimation that their child would never be able (anymore) to make any form of contact with them. The opposite also seemed to hold true. Parents often added that when they did not think their child was suffering, this made it hard for them to decide that treatment should be withheld or withdrawn. Moreover, if outcomes were still uncertain, they also found it hard to make this decision. Instead, they preferred treatment to be continued until there was more clarity about their child’s life expectancy and neurological prognosis.

The observational study shows that parents are well aware that their decisions are also influenced by their personal interests and by the interests of their family as a whole. They often tried to give words to the dilemmas this caused. For instance, several parents described that they did not want to prolong their child’s suffering, but that they also wished - for their own sake - to have their child with them for some more time. Other parents described that they realized how difficult and burdensome the care for their now severely disabled child would be, but that they still wished to provide this
care as long as their child would not be in pain and as long as he or she would recognize them.
(Chapters 4 and 5)

4. How do parents wish to be involved in end-of-life decisions for their child?

Our review indicates that most (but not all) parents prefer an approach of shared decision-making. For parents this concretely implies that they are honestly and emphatically informed about the remaining treatment options and about their child’s prognosis in a language they can understand and without being giving false hope. Moreover, that they get the opportunity to express what they see is happening with their child and what they think is now best to do. In that respect, parents also highly value to hear the physicians’ personal preferences. Finally, that they see that their viewpoints are being respected and integrated in the final decision to which they can (then) fully agree.22, 43, 45, 49-51, 53-58 A further analysis of the review studies indicates that some parents wish to share in all decision-making phases, yet leave the final say to their physicians. Other parents wish to make the final decision, in close consultation with their physician.22, 43, 45, 49-51, 53-58 It emphasizes the thin line between a shared approach and an informed approach in clinical practice.

The vast majority of studies included in the review were performed in Western, highly developed countries. The findings described above are therefore limited to these countries. More research is needed in developing countries before we can give a more complete answer to the question how parents in different cultures prefer to be involved in end-of-life decisions for their children.
(Chapter 4)

5. Why do conflicts arise between parents and physicians and within a team?

According to the Dutch pediatric specialists who participated in the interview study and in the survey, disagreements between parents and medical team arise regularly. This finding was confirmed in the observational study. Most parent-team conflicts arise because parents cannot yet agree to treatment discontinuation because of their strong religious convictions or because they have a more optimistic view of their child’s chance of survival or expected quality of life. These reasons may reinforce one another. Occasionally, conflicts arise because parents wish treatment to be discontinued against the opinion of the medical team. In these cases, parents do not think that treatment will be helpful for their child anymore, but by now only causes suffering. Our observational study shows that this type of conflict occurs more often than we previously assumed, based on the literature. When parents do not speak the physicians’ language fluently, this increases the risk of mutual misunderstanding and conflicts. Not only is it harder for parents to understand what is happening with their child, but concepts like suffering, futility and quality of life also have other connotations for them than for their physicians. These connotations are also influenced by parents’ religious beliefs.

Disagreements within medical teams occur less often than between parents and medical team. They are caused by one or more of the following factors: different views on the remaining treatment options; different ideas about the pace and carefulness of the decision-making process; different estimations of the chance of survival; and different opinions about the expected quality of life and what may be judged as sufficient quality of life.
(Chapters 2, 3, 5 and 6)

6. How are disagreements solved?

The main conflict solving strategy can be summarized in one word: time. When disagreements arise between parents and the medical team, physicians give parents more time to be with their child to say goodbye or (sometimes) to witness the changes in their child. Moreover, physicians take time to further speak with the parents, to arrange a second opinion in another medical center or to perform additional diagnostics. In solving disagreements with colleagues, physicians largely choose similar strategies: arranging further team discussions, doing additional diagnostics, asking a second opinion from colleagues elsewhere and taking time to await further developments or as it was often phrased: ‘letting the child show the way’. A last remedy to solve conflicts is asking the help of someone with authority or doing an appeal on authority. If parents’ religious convictions play a role, a clergyman whom the parents trust is asked to join the meetings. Sometimes, the physician proposes that he or she will now take over all responsibility for the decision, with parents’ agreement. Ongoing disagreements within a team are solved by giving the medical director the final say.
None of the cases in which there were conflicting views between parents and team (as reported in the survey and interview study and as observed in the observational study) were brought to court.
(Chapters 2, 3, 5 and 6)

7. How does physicians’ communication match with the parents’ communication in the conversations we observed?

All physicians extensively informed the parents about the child’s serious condition, the grave prognosis and lack of treatment options. Parents valued this, as became clear from their reaction and from their questions in return. These questions showed their understanding of the key issues. Physicians incorporated their preferences in the information they provided, for instance by emphasizing the child’s serious deterioration and the harms of treatment continuation. Although they were seldom invited to do so, almost all parents spontaneously presented their preferences regarding their child’s course of treatment. Two thirds of them emphasized that they did not prefer treatment to be continued at all cost. One third expressed a preference for treatment to be continued or restarted in contrast to the preference of the physicians. The majority of physicians presented the decision to withhold or withdraw treatment as a team decision for which parents could and should not be held responsible. Most parents expressed their agreement or assent. Some parents held on to their objections. In these cases the decision was postponed to give parents more time. In the following meeting parents were actively invited to present their points of view and to discuss their hopes and fears. Eventually, a decision was being reached to which all could agree.

Our combined quantitative and qualitative analysis leads to the conclusion that physicians’ and parents’ communication matched in the first decision-making stage of providing and receiving information. Yet in most cases, their communication did not fully match in the second stage of deliberating nor in the third stage in which a decision was being reached. In these stages it was clear that parents were able to handle a more active role than they were being given, despite their often intense emotions.
(Chapters 5 and 6)

8. Who should decide?

As we argue in-depth in chapter 6, parents should be given the opportunity to share in the decision whether their child’s treatment should be forgone. We therefore strongly recommend a shared approach which extends the current regular practice of asking parents for their agreement. The most impressive outcome of the observational study was witnessing the deep urge of all parents to protect their child from suffering. In all parents, this urge eventually outweighed their fear of losing their child. But only if parents get the opportunity to share in all three stages of the decision-making process, they can truly protect their child. Parents are not obliged to make use of this opportunity. Some parents may wish to leave the final decision to the physicians or they may not wish to participate in the decision-making process at all. Yet, this does not relieve physicians from the responsibility to inform parents about crucial developments and to check on a regular basis whether parents are feeling capable to share in the decision-making to a further extent (or not).
Parents may and should only be over-rulled as proxy decision-makers if they indisputably threaten their child’s best interests. This may occur if they refuse treatment that is clearly beneficial for their child or if they keep requesting futile treatment that will prolong the child’s suffering. Both situations occur rarely. They did not occur in our observational study and were not reported in the interview study nor in the survey.
(Chapters 2, 3, 5 and 6)
Putting our conclusions in a broader perspective

Because of the impact on all involved, end-of-life decision-making has become the subject of much research within neonatology and within pediatrics. This is clearly illustrated by the impressive amount of publications to which I referred at the beginning of this chapter, a list which is far from complete. In overlooking the studies published in the last 25 years, I would like to point out three interesting patterns.

First, that a considerable number of studies are ethical studies. They aim at clarifying much used and sometimes misused concepts like futility and suffering. They also aim at analyzing the values and interests which may conflict in making end-of-life decisions for a child.20, 24, 30, 31, 41, 50, 59-81 One conflict of interests has perhaps received most attention: that between the child's interests and the parents' interests. Authors like Diekema, Truog and Wilkinson have written in-depth about this specific dilemma.22, 41, 50, 63, 64, 67, 68, 70, 81, 82 They convincingly argue that the interests of the child and his or her parents may compete, but that this rarely happens to an extent that parents should be overruled as their child's proxy decision-makers. A study by Sharman as well as our observational study indicate that parents are often well aware of these conflicting interests.52 Moreover, they are able, and even welcome, to share this dilemma with their physicians. In our case report, we focused on a conflict of interests which has received less attention in the literature: that between the parents' interest to fully share in end-of-life decisions and their interest to be protected from too much responsibility and later doubts and regrets.

A second pattern is the growing number of studies investigating the role of parents in making end-of-life decisions and parents' experiences regarding this process. The way in which this is done varies. Several national and international studies have chosen an indirect approach by asking physicians how they involve parents in making end-of-life decisions and how they think parents should be involved. This type of study is mainly done by means of questionnaires. While earlier studies concluded that physicians from Northern European, North-American and Australian countries involved parents to a (far) greater extent than physicians working in Southern European and South-American countries, later studies indicate that these differences are starting to disappear. In most countries, it is now normal policy to inform and discuss crucial decisions with parents, although this does not automatically imply that parents also share in making these decisions.22, 26, 37, 82-91

Our review shows that less studies have directly investigated parents’ experiences and reflections. However, our inclusion period ended in 2010 and since then a considerable number of studies have been published in which parents were interviewed about the end-of-life decisions which had been made for their child, including the forgoing of artificial nutrition and hydration. These studies included relatively large groups of parents, which makes it possible to draw more general conclusions. All studies confirm the main outcome of our review: most parents highly value to fully share in the decision-making process, thereby receiving the emotional support of their physicians and nurses.21, 34, 40, 92-96 One study investigated parents’ long term grief in relation to their involvement in end-of-life decision-making and concluded that parents’ perception of a shared decision is associated with lower grief.92 All studies underline that individual preferences may vary and that parents should therefore get the opportunity to decide what role they wish to play.

Finally, it is remarkable that among the vast amount of studies investigating end-of-life decision-making in children, one category of studies is practically lacking: observational studies exploring the process of end-of-life decision-making in clinical practice. Our systematic search resulted in two studies, both within neonatology.50, 97 These studies gave an in-depth description of the complexity and delicacy of making decisions to withhold or withdraw treatment in neonates. However, they did not focus on how physicians and parents communicate about these decisions. In developing our observational study and our coding instrument we therefore gladly incorporated the methodological knowledge acquired in previous studies within adult care.17, 98, 99

Critical appraisal

What were the main strengths and limitations of our entire project? To answer these questions, I will shortly review the four successive studies we conducted. Our first study, the interview study among neonatologists, aimed to investigate the frequency and background of conflicts in neonatal end-of-life decisions. Eighty-five neonatologists were interviewed about 147 cases. These structured interviews, in combination with a review of the medical records, resulted in a reliable estimation of the frequency of these conflicts. Yet, the number of conflicts was small and this limited the generalizability of our conclusions about the main causes and risk factors. Furthermore, all findings were based on the physicians’ perceptions and not on the parents’ perceptions.
Our second study, the survey among 136 pediatric specialists, aimed to explore how physicians from different pediatric specialties reach end-of-life decisions, how they involve parents, and how they address conflicts. The high response rate made it possible to draw statistically reliable conclusions and provided new insights in how physicians involve parents in the decision-making process and in the factors influencing this approach. However, the outcomes of this study were also based on the physicians’ observations and opinions, not on the observations and opinions of the parents nor on real practice observations.

The fact that we did not know how parents experienced their involvement, motivated us to conduct our third study, the integrative review. This study aimed at systematically tracing and reviewing original studies that directly investigated parents’ perspectives. Our extensive search led to a relatively small number of studies, confirming that parents’ experiences have been less investigated than the experiences of physicians, at least up till then. The included studies varied in their conceptual clarity and methodological quality. This hindered a direct comparison, resulting in a limited generalizability of our outcomes. This was also due to the fact that most included studies had been conducted in Western countries.

The fourth, observational study was the most challenging and most time-consuming part of our project. It aimed at exploring how physicians and parents communicate about end-of-life decisions in clinical practice and at determining to what extent parents share in the decision-making process. Although we managed to include a large variation of cases, the total number of 19 cases was relatively low. Moreover, our study was limited to two University Medical Centers. At the same time, per case we observed all key-meetings and thoroughly analyzed the communication behaviors of the participating physicians and of the parents. This study completed our project.

In thinking about the strengths and limitations of the entire project, I see three main strengths. First, the phasing which enabled us to learn from the outcomes of a former study in designing the next one. Second, the fact that we managed to investigate the perspectives of the parents and the physicians and, eventually, their actual interaction. Finally, I think it is a strength that we used a rich body of medical, ethical, legal and psychological knowledge in designing our project and in interpreting our data. I also see three main limitations. First, despite the international character of the review, our other studies focused on the Dutch practice. Hence, it may be questioned how generalizable our findings are, also in light of the fact that the Netherlands have a more liberal policy than most other countries regarding end-of-life decisions like euthanasia and the active ending of life for newborns. But perhaps we can also conclude that our practice regarding parental involvement in end-of-life decision-making is less different than we thought. And also less progressive. A second limitation is that in all studies we focused on the perspectives of the physicians and the parents and not on the perspectives of the other professionals involved nor on other family members. We made this choice because physicians and parents are the main actors in making end-of-life decisions for a child and we therefore wished to start with studying their interaction by means of the interviews, survey and observational study. These phases took more time and effort than we had foreseen. We therefore had to abandon our original plan to include nurses, despite their important role in supporting parents. Third and finally, despite the fact that we thoroughly investigated the actual interaction between physicians and parents by means of our observational study, we did not investigate how all participants evaluated this interaction and what they found particularly helpful and what not. Our first idea was to interview parents and physicians shortly after each meeting. We abandoned this plan because of the risk that it would ask too much of the parents and because of the risk that it would influence the decision-making process. We then decided to interview the parents and the physicians at a later stage. I started with interviewing the parents of 7 included children at their home, 6 months after the last decision had been made. Because of my serious illness in 2012 and 2013, I could not continue these interviews. However, the most extensive interview with one couple of parents was the starting point for Chapter 6.

**Recommendations**

There is still much to be known about end-of-life decision-making in children and about how parents and physicians communicate about these decisions. Our research project leads to new ideas for further research, as research projects often do. Below, I will present these suggestions. Based on our conclusions, I will also formulate practical recommendations to improve a shared process of end-of-life decision-making in children. Some of them are meant for the parents, some for the physicians (and possibly for other professionals involved) and some for hospital boards. I will start with these do’s and don’ts.
### Practical recommendations

#### Recommendations for parents:

1. Write down any questions regarding your child’s situation, treatment and impending decisions. Tell the physician or the nurse who is coming with you that you would like to discuss these.
2. Share your observations and worries regarding pain and other symptoms of suffering and ask how they are or will be treated.
3. Ask for a written summary of what has been discussed, if you think this is helpful.
4. Consult other experts or other parents if you think this is helpful and ask your physician to help you to arrange this consultation.
5. Ask for one physician as spokesman, especially if more pediatric specialists are involved in your child’s care and if their information and advice starts to differ.

#### Recommendations for the medical team:

1. Invite parents to present their observations, views and preferences regarding their child’s treatment in each meeting without pushing them to do so.
2. Concretize what you mean by terms like medical futility, unbearable suffering and (no) quality of life.
3. Give parents the opportunity to fully share in making decisions to continue or discontinue treatment on behalf of their child.
4. Be conscious of your personal experiences, values and emotions and how they influence your decision-making. Be honest about them to colleagues and, if you consider this helpful, also to parents.
5. Welcome divergent points of view from parents and colleagues. Do not act defensive.
6. Avoid speaking of decisions if there is nothing to decide yet or nothing to decide anymore.

#### Recommendations for hospital boards:

1. Invest in sufficient time for medical teams to timely discuss end-of-life decisions with patients or their proxy decision-makers.
2. Employ physicians, nurses and social workers from culturally and linguistically diverse backgrounds. Provide good access to chaplains from different religions and to translators.
3. Invest in inter-collegial consultation and coaching and in training of advanced communication skills.
4. Create a safe atmosphere in which it is normal to share good and bad experiences regarding difficult conversations within medical teams without repercussions.

#### Recommendations for future research:

1. Investigate in Western and especially in non-Western countries how end-of-life decisions are being discussed with parents and how this communication is being valued by the parents, by their physicians and by others being present. Especially look at the phrasing of much used concepts like futility, suffering and quality of life.
2. Study the long term effects of conflicts in end-of-life decision-making on parents’ and on professionals’ well-being.
3. Explore how parents’ and physicians’ concepts about quality of life influence their treatment decisions in children who are or might become severely impaired.
4. Further investigate the influence of parents’ and physicians’ beliefs and emotions on their decision-making and on their coping.
5. Investigate how end-of-life decisions are being discussed with children of different age groups and what they value as helpful in this communication.
Closing statement

Not so long ago a social scientist I had not met before made an interesting remark. She said that researchers should avoid investigating dilemmas which they had experienced in their personal lives. These experiences and their accompanying emotions and convictions would seriously hinder good, objective research. Her remark reminded me of the discussion among physicians whether their personal emotions are hindering them in making good clinical decisions. I already argued that this does not need to be the case. If professionals find the right balance between honestly acknowledging their emotions and not being carried away by them, their clinical decision-making will most probably improve because they better understand what is at stake for the child, the parents and for themselves. Moreover, their communication will be more empathic and genuine which may prevent conflicts from escalating. It may even be postulated that physicians who cope with their emotions instead of blocking them off, have less risk of developing burnout symptoms.100-103

But what about researchers? In contrast to physicians they can decide to not take on a specific project (although this freedom also has its limits). Should they? For some time I thought they should or, to be more precise, that I should. Fate or bad luck: some lives are less easy than others. My life was not easy after the loss of our son and after our daughter was diagnosed with Angelman Syndrome, two years later. Despite my grief I could not prevent myself from observing and questioning what went on. Why had the team of neonatologists advised us to withdraw treatment when there was still a chance our son could survive but with very severe disabilities? Why had the pediatrician never raised the subject of treatment limitation when it was 100% sure that our daughter had indeed very severe disabilities? What made that we first fully complied with the advice of the neonatologists and then with the approach of the pediatrician who was clearly fond of our daughter? And was it not her fondness that helped us to see our daughter’s life as a life ‘worth living’? Or was this view merely our way of ‘coping with the uncopable’ as a psychologist once explained? I kept making notes of these thoughts and left it at that. After our two other children were born, we regained our balance. In the eyes of others we remained a special family, but in our own eyes we became a quite normal, happy family. And then, after I had started a new job at the Emma Children’s Hospital AMC, I met Tom’s mother. In the months to come I spoke with more parents as well as physicians and nurses. They all felt that more knowledge was needed to improve the process of end-of-life decision-making and parents’ involvement in this process. This need persuaded me to resume my work as a researcher. Yet, with two safeguards. I asked my three supervisors and a broader group of advisors to critically evaluate my data collection and analysis. Moreover, I asked the help of a coach when I began observing the end-of-life conversations between parents and physicians. With this coach I primarily discussed how to deal with the most difficult part of this phase: observing how conflicts arise and not being able to do something about it because as a researcher you are there to watch and not (yet) to advise.

By now, I no longer think that researchers should avoid investigating dilemmas which may cause emotions due to personal experiences. On the contrary: I think that researchers with personal knowledge of the hard sides of life will often be more sensitive researchers, on the condition that they ask the help of others to watch over them and on the condition that they are truly interested in the topic they wish to study.
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Appendix
Summary

End-of-life decisions are among the most difficult decisions to make, from a medical, ethical, legal and psychological point of view. They often confront us with the limits of our medical knowledge, with moral dilemmas without best answers, and with feelings of powerlessness, injustice and grief. Making end-of-life decisions for a child is an even more burdensome process. There is a substantial difference between deciding to forgo treatment together with an adult patient who can look back on a life worth living or making this same decision with parents for a child whose life has only just begun. The most difficult part of making this decision is that it concerns a life and death decision for another human being. Moreover, in many cases we will never be 100% sure that this decision was the best for this particular child and that this moment was the best moment to do so.

Because of the impact on all involved, end-of-life decision-making has become the subject of much research within neonatology and within pediatrics. These studies have focused on different aspects of this complex process. Most studies conducted in the last 25 years, quantitatively investigated the neonatal and pediatric practices of end-of-life decision-making in and over countries, with special attention for the number and types of end-of-life decisions in relation to the child’s dying process. Furthermore, a considerable amount of studies explored the attitudes and opinions of physicians and nurses regarding end-of-life decision-making in neonates and children. Few studies looked into the experiences and perceptions of parents facing end-of-life decisions for their child, while studies investigating how physicians and parents actually communicate about these decisions are largely lacking.
Yet, there is growing consensus that physicians should tailor their decision-making approach to parents’ preferences and needs. But how can physicians best do this? Should they ask parents what extent they wish to be involved in the decision-making process? How extensively should they inform them about all concerns and remaining options? Can parents be asked what they think is best to do? And what to do if parents and physicians start to have conflicting points of view on treatment continuation or discontinuation for a child? These questions were the starting point for our research project ‘Sharing the burden of deciding. How physicians and parents make end-of-life decisions’. We describe the outline of this project in chapter 1, after having introduced our ethical and legal framework. Between 2008 and 2015 we performed four successive studies: an interview-study among Dutch neonatologists; a survey among Dutch pediatric intensivists, oncologists, neurologists, neurosurgeons and metabolic pediatricians by means of a questionnaire; an integrative review of international empirical studies among parents; and an observational study of end-of-life conversations between parents and physicians in two University Medical Centers. These outcomes are described in the following chapters.

Chapter 2 presents the results of a national cross-sectional survey among pediatric intensivists, oncologists, neurologists, neurosurgeons, and metabolic pediatricians practicing in the 8 Dutch university hospitals. By means of a questionnaire we investigated how these pediatric specialists reach end-of-life decisions, how they involve parents in the decision-making process and how they address conflicts. Of the 185 eligible pediatric specialists, 74% returned the questionnaire. All responding physicians generally discuss an end-of-life decision with colleagues before discussing it with parents. In half of the reported cases, the respondents informed parents about the intended decision and asked their permission to act accordingly. In one-quarter of the cases, the respondents informed parents without asking for their permission. In the remaining one quarter of the cases, the respondents informed and advised parents and consequently allowed them to have the decisive voice. The approach physicians chose in the reported cases was not significantly influenced by personal characteristics like their gender, years of experience and religious affiliation. Instead, we did find that this approach was highly influenced by the type of decision being considered, the type of treatment the child received and the length of the relationship between the physician and the family. For instance, physicians facing a decision to withhold life-sustaining treatment in a child receiving palliative terminal care tended to give their advice and then let parents make the final decision, while physicians facing a decision to withdraw life-sustaining treatment (mainly mechanical ventilation) tended to protect parents from making this ‘active and technical’ decision. Yet, they strived to get parents’ agreement or at least their assent. Sometimes, conflicts within medical teams arose as a result of uncertainties about prognosis and treatment options. Most conflicts with parents arose because parents had a more positive view of the prognosis or had religious objections to treatment discontinuation. All conflicts were eventually resolved by a combination of strategies. Main conflict-solving strategies were taking more time and extending discussions.

Chapter 3 focuses on the frequency and background of conflicts about neonatal end-of-life decisions. The medical files of 359 newborns who had died during one year in the 10 Dutch neonatal intensive care units were reviewed, resulting in the identification of 150 deaths that had been preceded by an end-of-life decision based on the child’s poor prognosis. The attending neonatologists of 147 of these 150 newborns were interviewed to obtain details about the decision-making process. End-of-life decisions about infants with a poor prognosis were mainly initiated by the physician, who subsequently involved the parents. Conflicts between parents and the medical team occurred in 18 of 147 cases and were mostly about the child’s poor neurologic prognosis. Conflicts within the team occurred in 6 of 147 cases and concerned the uncertainty of the prognosis. In the event of conflict, the end-of-life decision was postponed. Consensus was reached by calling additional meetings, performing additional diagnostic tests, or obtaining a second opinion. The chief causes of conflict encountered by the physicians, were parents’ religious convictions that forbade withdrawal of life-sustaining treatment and poor communication between the parents and the team.

In Chapter 4, we describe the outcomes of an integrative review. In PubMed, EMBASE, CINAHL and PsycInfo, we systematically searched for qualitative and quantitative original studies that directly investigated the experiences of parents of children between 0 and 18 years of age for whom a decision to withhold or withdraw life sustaining treatment had been made. We identified 17 studies that met all inclusion criteria. Parents reported a wide variation in how they were involved in end-of-life decisions for their child, ranging from having no involvement to being the sole decision maker. The majority of parents preferred to actively share in decisions to withhold or withdraw life sustaining treatment. This concretely implied that they wished to be honestly informed, to be heard and respected in their views and to be asked for their agreement. For some parents, it furthermore implied that they wanted to make the final decision together with their physicians.
Other parents did not wish to bear this responsibility, especially in cases with considerable uncertainties regarding prognosis. Main factors which influenced parents in their decision-making were: the visible suffering of their child, the fact that there was no chance of survival anymore or the prognosis of a very poor quality of life.

Chapter 5 presents the outcomes of a prospective observational study which we conducted in two Dutch University Medical Centers. This study aimed at exploring how physicians communicate with parents about end-of-life decisions for their child and to what extent parents share in the decision-making process. Overall, 27 physicians participated and 37 parents of 19 children for whom a decision to withhold or withdraw LST was being considered. Forty-seven conversations were audio-recorded, ranging from 1 to 8 meetings per child. By means of a coding instrument, we quantitatively and qualitatively analyzed physicians’ and parents’ communication. On average, physicians spoke 67% of the time, parents 30% and nurses 3%. All physicians focused primarily on providing medical information, explaining their preferred course of action and informing parents about the decision being reached within the team. In only 2 cases, parents were asked to share in the decision-making. Despite their intense emotions, most parents took considerable effort to actively participate in the conversation. They did this by asking for clarifications, offering their preferences and reacting to the decision being proposed (mostly by expressing their assent). In the few cases where parents strongly preferred life sustaining treatment to be continued, the physicians either gave parents more time or revised the decision. Parents’ greatest concern was that their child might suffer in the phase of dying. We conclude that parents are able to handle a more active role than they are currently being given.

In Chapter 6, we present the case of a 3-year-old boy with acquired brain damage due to autoimmune mediated encephalitis whose parents wanted treatment to be stopped against the views of the medical team. By comparing this case with relevant literature, we systematically explore the pros and cons of sharing end-of-life decisions with parents in cases where treatment is considered futile by parents and not (yet) by physicians. We conclude that sharing end-of-life decisions with parents is a more important duty for physicians than protecting parents from guilt or doubt. Moreover, a request from parents on behalf of their child to discontinue treatment is, and should be, hard to overrule in cases with significant prognostic uncertainty and/or in cases with divergent opinions within the medical team.

In Chapter 7, the general discussion, we return to the eight questions which were the starting point for this research project. Based on a synthesis of the individual study outcomes, we formulate overall answers to these questions and discuss the main strengths and limitations of this project as a whole. Finally, we formulate ideas for further research and present practical recommendations to improve a shared process of end-of-life decision-making in children. Some of these recommendations are meant for the parents, some for the professionals involved and some for hospital boards. With a personal statement I finally close this project.
Samenvatting

In Nederland overlijden jaarlijks ruim 1000 kinderen tussen 0 en 16 jaar. Een groot aantal van hen stervt op een neonatale of kinder intensive care afdeling. Aan hun overlijden gaat in veel gevallen de beslissing vooraf om de behandeling te staken of niet opnieuw te starten. Deze beslissing wordt genomen in situaties waarin een kind ondanks alle hoog-technische zorg geen levenskansen meer heeft. Soms is er nog wel een (zeer) geringe kans op overleven maar lijdt het kind onder de zwaarte van het behandelingstraject zonder dat er nog enig uitzicht is op een minimale kwaliteit van leven, met andere woorden op een leven ‘waard om geleefd te worden’. Ook dan kan het medisch team in nauw overleg met ouders besluiten om af te zien van verdere behandeling zodat het kind zo vreedzaam mogelijk kan overlijden. Het is duidelijk dat einde-van-leven beslissingen een zware wissel trekken op alle zorgverleners die nauw bij het kind betrokken zijn, in de allereerste plaats de ouders.

Ook binnen de volwassen-geneeskunde gaat aan iedere beslissing om af te zien van verdere behandeling een ingrijpend proces vooraf. Toch maakt het een groot verschil om samen met een patiënt die terug kan kijken op een rijk en vol leven te beslissen over het staken van de behandeling of om deze beslissing te moeten nemen voor een kind dat nog maar net aan zijn of haar leven is begonnen. In het eerste geval zal er eerder sprake zijn van berusting en acceptatie. In het tweede geval overheersen gevoelens van onrechtvaardigheid, onmacht en diep verdriet. Er is nog een ander substantieel verschil. In het eerste geval neemt de patiënt zelf de uiteindelijke beslissing, dit in nauwe samenspraak met zijn of haar arts. In het tweede geval beslissen artsen in nauwe samenspraak met ouders over leven en dood van een kind dat daartoe zelf niet (meer) in staat is. Deze
hoofdstuk 1

kinderoncologen, kinderneurologen, neurochirurgen en metabole neonatologen; een survey onder Nederlandse kinderarts-intensivisten, verschillende onderzoeksfasen uit: een interview-studie onder Nederlandse van-leven beslissingen plaatsvinden. Tussen 2008 en 2015 voerden we vier de ethische en juridische kaders te hebben toegelicht waarbinnen einde-van-leven beslissingen te mogen nemen en hoe zij denken over de rol van ouders. Een kleiner aantal studies exploreerden de ervaringen van ouders zelf nadat zij geconfronteerd waren met een einde-van-leven beslissing voor hun kind. Tot slot blijkt een type onderzoek nagenoeg te ontbreken: observationele studies naar hoe einde-van-leven beslissingen in de dagelijkse praktijk tot stand komen en naar hoe artsen en ouders over deze beslissingen communiceren.

Uit de studies die tot nu toe gedaan zijn blijkt dat artsen de manier waarop zij ouders betrekken bij einde-van-leven beslissingen het liefst afstemmen op de voorkeur en op de draagkracht van het individuele ouders met wie zij om tafel zitten. Tegelijkertijd vragen zij zich af hoe ze dit concreet moeten doen. Kunnen zij ouders op de man af vragen in welke mate zij betrokken willen worden bij het besluitvormingsproces? Een besluitvormingsproces dat naar alle waarschijnlijkheid zal leiden tot het versneld overlijden van hun kind? Hoe gedetailleerd moeten zij ouders informeren over de prognose van hun kind, over de overgebleven behandellopties en de voorziene beslissingen? Kan aan ouders wel gevraagd worden welke beslissing zij het meest in het belang van hun kind vinden? Zullen zij niet altijd kiezen voor het voortzetten van de behandeling? En tot slot: als er dan verschillen van inzicht ontstaan tussen artsen en ouders, hoe kunnen deze het beste worden overbrugd?

Deze vragen vormden het startpunt voor het onderzoeksproject ‘Sharing the burden of deciding. How physicians and parents make end-of-life decisions’. In hoofdstuk 1 beschrijven we de opzet van dit project, na eerst de ethische en juridische kaders te hebben toegelicht waarbinnen einde-van-leven beslissingen plaatsvinden. Tussen 2008 en 2015 voerden we vier verschillende onderzoeksfasen uit: een interview-studie onder Nederlandse neonatologen; een survey onder Nederlandse kinderarts-intensivisten, kinder-oncologen, kinderneurologen, neurochirurgen en metabolie kinderartsen; een review van alle empirische studies die wereldwijd zijn gedaan naar de ervaringen van ouders zelf; en een exploratieve studie waarin we einde-van-leven besprekken tussen ouders en artsen in twee Nederlandse Universitaire Medische Centra observeerden en analyseerden. De uitkomsten uit deze vier onderzoeksfasen belichten we in de verdere hoofdstukken.

In hoofdstuk 2 presenteren we de resultaten van de landelijke survey onder kinderartsen, werkzaam in één van de 8 Nederlandse Universitaire Medische Centra. Door middel van een vragenlijst onderzochten we hoe zij komen tot een einde-van-leven beslissing, hoe zij ouders bij deze beslissing betrekken en hoe zij omgaan met eventuele conflicten die tijdens dit proces ontstaan. Van de 185 aangeschreven artsen retourneerde 74% de vragenlijst. Al deze artsen gaven aan dat zij een einde-van-leven beslissing liefst eerst met collega’s bespreken voordat zij met ouders in gesprek gaan. Gevraagd naar de meest recente casus waarin zij een einde-van-leven beslissing hadden genomen, bleek dat in de helft van deze gevallen de betreffende artsen de ouders hadden geïnformeerd over de voorgenoemde beslissing en hen vervolgens hadden gevraagd of zij met deze beslissing konden instemmen. In een kwart van de gevallen hadden de artsen de ouders alleen geïnformeerd over de voorgenoemde beslissing. In de overgebleven gevallen hadden de artsen ouders hun advies gegeven en de uiteindelijke beslissing aan hen gelaten. Er bleek geen significant verband te bestaan tussen de benadering die artsen hadden gekozen en persoonlijke karakteristieken zoals hun geslacht, aantal ervaringsjaren of religieuze overtuiging. In plaats daarvan bleek de gekozen benadering significant samen te hangen met de soort beslissing die werd overwogen, de aard van de behandeling die het kind kreeg en de duur van de behandelingrelatie tussen de arts en het kind. Zo waren artsen die de beslissing overwogen om geen levensondersteunende behandeling (meer) te starten bij een kind dat na jaren van curatieve behandeling nu alleen nog palliatieve zorg ontving het meest geneigd om ouders de uiteindelijke beslissing te laten nemen. Dit in tegenstelling tot artsen die de beslissing overwogen om een levensondersteunende behandeling te staken, met name beademing. Zij waren het meest geneigd om ouders te beschermen tegen het nemen van deze ‘actieve, hoog-technische’ beslissing. Tegelijkertijd wilden zij wel graag weten of ouders met deze beslissing konden instemmen. De meeste verschillen van inzicht tussen artsen en ouders ontstonden omdat de ouders het idee hadden dat hun kind nog zou kunnen herstellen en/of doordat ouders op grond van hun religieuze gay selafschriften hadden tegen het staken van de behandeling. Alle verschillen van inzicht werden uiteindelijk opgelost door
een combinatie van strategieën, met als belangrijkste het nemen van meer tijd en het uitbreiden van het aantal overleggen.

In hoofdstuk 3 rapporteren we over het aantal conflicten rond einde-van-leven beslissingen binnen de neonatologie en over hun belangrijkste oorzaken en oplossingsstrategieën. De medische statussen van 359 pasgeboren die binnen een periode van een jaar waren overleden op een van de 10 Nederlandse neonatale intensive care afdelingen werden doorgelicht. Dit resulteerde in de identificatie van 150 sterfgevallen die waren voorafgegaan door een einde-van-leven beslissing, gebaseerd op de zeer slechte neurologische prognose van het kind. De neonatologen die betrokken waren geweest bij de zorg voor 147 van deze 150 pasgeboren werden geïnterviewd. In 18 van de 147 casus bleken er conflicten te zijn ontstaan tussen ouders en het medisch team. Deze werden meestal veroorzaakt door verschillen in inzicht over de neurologische prognose van het kind. Gewetensbezwaren van ouders op grond van hun religieuze overtuiging en moeiemate communicatie tussen artsen en ouders droegen bij aan het ontstaan van deze verschillen in inzicht. In 6 van de 147 casus ontstonden er verschillen van inzicht binnen het medisch team, met name doordat er onzekerheid was over de neurologische prognose. In alle casus werden de verschillen in inzicht uiteindelijk opgelost doordat de beslissing werd uitgesteld en er in die tussentijd extra werd overlegd, er aanvullende diagnostiek werd gedaan en/of een second opinion werd ingewonnen.

In hoofdstuk 4 beschrijven we de uitkomsten van de systematische literatuurstudie. Binnen Pubmed, EMBASE, CINAHL en PsyCInfo zochten we naar kwalitatieve en kwantitatieve studies waarin ouders zelf zijn gehoord over hun ervaringen rond de einde-van-leven beslissingen die genomen werden voor hun kind. Uiteindelijk konden we 17 studies includeren. Uit een onderlinge vergelijking van deze studies blijkt dat ouders wereldwijd heel verschillend worden betrokken bij beslissingen over de behandelwijze van hun kind te staken of niet meer in te stellen. Dit varieert van geen enkele inbreng hebben tot de uiteindelijk beslissing alleen moeten nemen. Gevraagd naar hun wensen gaven de meeste ouders aan dat zij het liefst actief wilden participeren in het besluitvormingsproces (of dit in het nazien hadden willen doen). Concreet hiedt dit in dat zij graag eerlijk en volledig wilden worden geïnformeerd over de prognose en resterende behandelopties, dat zij op hun beurt hun observaties en visies wilden delen en dat zij de gelegenheid wilden krijgen om al dan niet in te stemmen met het uiteindelijke besluit. Sommige ouders gingen nog een stap verder en gaven aan dat zij ook graag de uiteindelijk beslissing wilden nemen, samen met hun arts. Andere ouders wilden liever dat hun arts de uiteindelijke beslissing nam, zeker wanneer er nog onzekerheid bestond over de prognose van hun kind. In hun besluitvorming voelden ouders zich vooral beïnvloed door de volgende factoren: het zichtbare lijden van hun kind; de (on)zekerheid over zijn of haar overlijden; een zeer slechte prognose dat betreft kwaliteit van leven.

In hoofdstuk 5 doen we verslag van de uitkomsten uit de prospectieve observationele studie in twee Universitaire Medische Centra. In deze studie verkenden we hoe artsen met ouders communiceren over einde-van-leven beslissingen en in welke mate ouders participeren in het besluitvormingsproces. Zevenentwintig artsen en 37 ouders van 19 kinderen waren bereid om deelnemers aan deze studie. In totaal konden we 47 gesprekken op band opnemen, variërend van 1 tot 8 gesprekken per kind. De gesprekken werden zowel kwalitatief als kwantitatief geanalyseerd door middel van een nieuw ontwikkeld codeerinstrument. Gemiddeld spraken artsen 67% van de tijd, ouders 30% en verpleegkundigen 3%. Vrijwel alle artsen besteedden het grootste gedeelte van hun spreektijd aan het geven van medisch-technische informatie, het voorbereiden van ouders op de voorziene beslissing en, uiteindelijk, het informeren van ouders over de einde-van-leven beslissing die binnen het team was genomen. In 2 van de 19 casus gaven de artsen de beide ouderparen de gelegenheid om mee te beslissen. Ondanks hun intense emoties deden de meeste ouders veel moeite om actief bij te dragen aan het gesprek. Zo namen zij zelf het initiatief om verhelderende vragen te stellen, om hun wensen wat betreft het verdere behandelsbeleid toe te lichten en om te reageren op de beslissing die hen werd voorgelegd (meestal door aan te geven dat zij zich hierin konden vinden).

In het kleine aantal gevallen waarin ouders wilden dat de behandeling werd gecontinueerd, gaven de artsen hen meer tijd om toe te groeien naar de beslissing of herzagen zij de beslissing. De grootste angst van alle ouders was dat hun kind zou lijden tijdens het terugtrekken van de behandeling en het stervensproces dat daarop zou volgen. We concluderen dat ouders in staat zijn tot een actiever aandeel aan het besluitvormingsproces dan artsen hen op dit moment geven.

Hoofdstuk 6 beginnen we met een diepgaande beschrijving van het besluitvormingsproces rond een jongetje van 3 jaar met verworven neurologische schade na een doorgemaakte encefalitis. In deze casus wilden de ouders dat de behandeling van hun kind werd gestopt, dit in tegenstelling tot het medisch team. In het vervolg van dit hoofdstuk vergelijken we deze casus met relevante literatuur en exploreren de argumenten voor en tegen
het gezamenlijk nemen van een einde-van-leven beslissing, met name in het geval dat ouders verdere behandeling medisch zinloos vinden en het medisch team (nog) niet. We concluderen dat het gezamenlijk nemen van einde-van-leven beslissingen een belangrijker plicht voor artsen is dan het beschermen van ouders tegen mogelijke gevoelens van schuld en twijfel. Verder beargumenteren we dat het verzoek van ouders om af te zien van de verdere behandeling van hun kind niet terzijde kan worden geschoven wanneer er prognostische onzekerheden zijn en/of verschillen van inzicht bestaan binnen het medisch team.

In hoofdstuk 7, de algemene discussie, keer ik terug naar de acht vragen waarmee we dit onderzoeksproject begonnen. Gebaseerd op een synthese van de uitkomsten uit de afzonderlijke onderzoeksfasen, formuleer ik overkoepelende antwoorden op deze vragen en bespreek ik de beperkingen en de sterke punten van het onderzoeksproject als geheel. Tot slot volgen ideeën voor verder onderzoek en praktische aanbevelingen hoe te komen tot meer gedeelde besluitvorming rond einde-van-leven beslissingen bij kinderen. Sommige aanbevelingen zijn bedoeld voor ouders, sommige voor artsen (en andere zorgverleners), sommige voor de Raden van Bestuur van een ziekenhuis. Met een persoonlijk postscriptum rond ik dit proefschrift uiteindelijk af.
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Curriculum vitae

Mirjam de Vos is a researcher in the field of medical ethics and medical communication. She works in the Academic Medical Centre (AMC) of Amsterdam. She combines her research with working as an ethical consultant and as a trainer in communication skills for the Department of Pediatrics. Besides her work in the AMC, she is senior consultant for the Dutch Centre of Consultancy and Expertise.

Mirjam was born in 1960 and studied child psychology and pedagogics at the University of Leiden. During her study she worked as a family guardian on behalf of the Council for Child Protection. In 1986, she graduated cum laude on a research project investigating suicidal behaviour in youth living in Dutch residential care homes. From 1986 until 1991, she worked as a child psychologist at the pediatric oncology department of the AMC. After having completed a postgraduate course in hypnosis, she developed a training programme in pain- and stress reduction techniques for children. In 1991, she started working as a free-lance trainer and consultant in pain management for the Dutch University Medical Centres.

In this same period, Mirjam’s eldest daughter was diagnosed with severe cognitive and motor disabilities. Conversations with medical specialists and nurses involved in the care for her daughter resulted in the handbook ‘Special care for special children’ (‘Bijzondere zorg voor bijzondere kinderen’). Physicians, nurses, paramedics and psychosocial workers with special expertise contributed to this handbook. It was published in 2000 by Elsevier.

In 2001, she returned to the AMC with the special assignment to improve the hospital care for severely disabled children. By then she had finished a post-graduate course in Medical Ethics at the University of Leiden. In 2008, she got funding for a research project entitled ‘End-of-life decision making in pediatrics’ by the Netherlands Organisation for Health Research and Development. This project consisted of several research phases and resulted in her thesis. From 2012 until 2014, Mirjam combined her research with working as a senior advisor for the Centre for Ethics and Health of the Dutch Ministry of Health, Welfare and Sport. On behalf of this organisation she wrote a book of interviews with patients and medical specialists: ‘When sharing decisions is no common part of medical practice’ (‘Wanneer samen beslissen niet vanzelf spreekt’). It was published in 2014.
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Sharing the burden of deciding
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