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At the boundaries of life

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CHAPTER 10

General discussion

Father: I think that our physicians will tell us, will recognize the moment when we have to stop treating.

Mother: For us as parents, I think it is impossible to recognize that moment, so we discussed that with our physicians. I have asked them: ‘when the moment is there, tell us. Because everything inside us tells us... that we shouldn’t do it, because it is our child. (...) We said: we will trust your advice in that.’

Father: We are very lucky to have physicians who are willing to do that, because from other parents we hear that many physicians are not willing to take that step.

(...)

Father: Our physicians have always said: ‘doing nothing can also be a way of doing something for your child.’ Parents will always doubt, because their rationale and their heart clash. But when a physician can assure you that by doing nothing, you can also do something for child, your head and heart unite a bit. So you don’t have to feel guilty, because I think that a lot of parents feel guilty when thinking about [life-limiting treatment]. So when a physician tells you: “You don’t have to look at your child and.. by doing nothing, you are doing so much for him.” That is such an important message to hear.

1. Introduction

When talking about life threatening conditions in young children, the hope and future that come with a young life meet the possibility of death. In this situation, the usual course of action is to provide curative, life prolonging and/or palliative care to increase the child's chances and relieve suffering.^{1,2,3} In medicine, physicians still tend to err on the side of life. As Clark describes:

“Palliative care has encouraged medicine to be gentler in its acceptance of death, yet medical services in general continue to regard death as something to be resisted, postponed, or to be avoided.”⁴

Despite this disposition, there are situations in medicine where decisions are made that aim at accepting death. Decisions of this kind are extremely difficult decisions to make, not only because death is a topic that seems to create discomfort in medicine,⁵⁻⁷ but also because in paediatrics decision-making often concerns children who are not able to fully participate in the decision-making process. It seems that in medicine very few decisions are as difficult as the decision to let a child die.⁸⁻¹⁷ However, during this research I interviewed a paediatrician who nuanced that view for me. During our interview the participant talked about the difficulties of making life-shortening decisions, and said the following:

“People often accuse me of sitting on God's chair when I discuss end-of-life decisions, or when I decide to limit treatment. But that is quite unfair, because I am sitting on God's chair every day, by making the decisions that keep these children artificially alive. Without knowing if that is what they want, whether I am not prolonging their unbearable suffering without them being able to tell me so.” Can someone tell me why I am being accused of sitting in God's chair if I'm proposing to stop that artificial prolongation, and I'm not accused of it when I keep them alive?
(interview P25)

Deciding to let a child die is an immensely difficult decision to make, but in a way prolonging a life can be too. This quote highlights why the strict separation between end-of-life decisions and life-prolonging care is perhaps sometimes unnatural, especially in paediatrics where the children are so often dependent on the life-prolonging care that is provided by physicians. The

decision to let a child live can be a difficult and courageous decision to make as well. In a way, this is the main message of this thesis: end-of-life decisions are as much about living as they are about dying.

To investigate the decisions and care that navigate the field between life and death, we conducted a nation-wide qualitative research. Our aim was to answer the following research questions:

1. What is the current context of regulations on care and decision-making in children with life-threatening conditions?
2. What does ‘good care’ mean for children who face a possibility of dying?
3. What does suffering mean in children with life-threatening conditions?
4. What are perceptions of parents and physicians on end-of-life decision-making?
5. Do parents and physicians perceive a need for regulations on active life-ending?

This final chapter presents and debates the conclusions to these questions. But first, a closer examination of the methodological considerations in this thesis is needed, including the question if, and how children should have been included in the study design. Next, the conclusions to each of our research questions will be presented. In the final part of this chapter, the conclusions are placed in the context of political and practical developments in the field of paediatric palliative care and end-of-life regulations. Finally, we discuss what the possible implications for future research are.

2. Methodological considerations

2a. Methodology and strengths

In 2015 the Dutch minister of Health, Welfare and Sports requested a large research project into care and decision-making around the end of life of young children.¹⁸ To fully explore the context of care and decisions, our research group conducted a research project containing three phases: a qualitative death-rate study, a qualitative interview study, and a questionnaire based on the outcomes of the qualitative study (see figure 1).

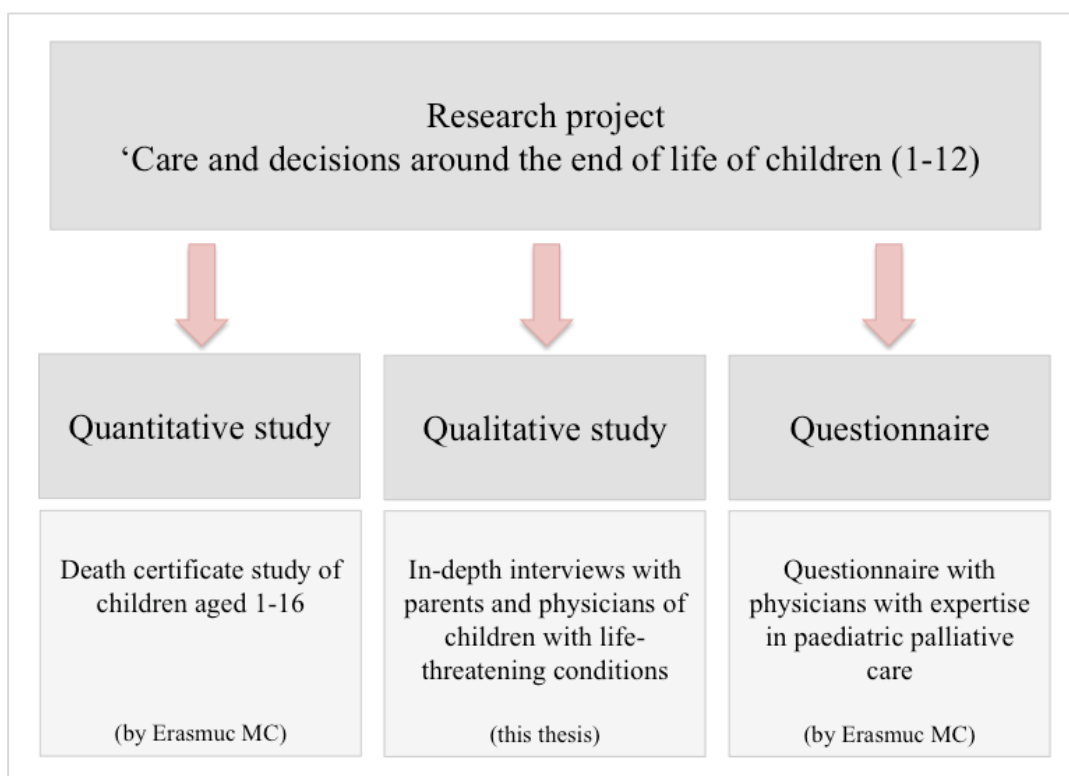


Figure 1. Study outline of research project ‘Care and decisions around the end of life of children 1-12.’

In the quantitative study project –a death certificate study— researchers from the Erasmus Medical Centre in Rotterdam explored the rate of life-limiting decisions that precede the death of a child (1-16years). Although the results are so far unpublished, the study provides much needed insights into the frequency of end-of-life decisions in paediatrics, including the result that there is no evidence that active life-ending is currently being practiced for children in this age group.¹⁹ Following our qualitative study, a questionnaire was used to check the findings of the qualitative study in a larger group of experts in paediatric palliative care.¹⁹ The qualitative study forms the main body of this thesis. The results of these three studies have led to a research report with conclusions and recommendations on care, decision-making and active life-ending in children with life-threatening conditions (aged 1-12). In September 2019, the results were presented to the minister of Health, Welfare and Sport and to the broader public.¹⁹⁻²²

While the death certificate study gives insight into quantitative data surrounding end-of-life decisions, such as the frequency of end-of-life decisions, it did not give insight into the circumstances and motivations behind the decisions. Such questions need a qualitative approach. In the studies in this thesis the different backgrounds, considerations and assumptions and that underlie such decisions are explored. The results give insight into a rich and diverse palette of experiences in paediatric palliative care and decision-making. Especially in palliative care, where existential themes play an important role (i.e. suffering, quality of life, perceptions on dying)^{2,23} qualitative research is a crucial tool to gain scientific insights.²³⁻²⁵

Performing a qualitative study comes with certain considerations. It has often been argued that qualitative studies are not able to reach the aim of scientific objectivity.²⁵ Qualitative research is a process of constant interpretation, from the moment of recruitment until data analysis. This means that in qualitative research –not unlike, as many might argue, quantitative research—^{26,27} has to be careful about its claims of objectivity.^{25,28} Analysing another human being's language requires researchers to interpret, and the collected data is inevitably influenced by the researcher's own perspectives and interactions with the participant.^{24,25,28} Therefore, it is important to be transparent about where we are subjective, and furthermore, incorporate as many perspectives as possible, to transition from a subjective perspective to a shared, intersubjective perspective.

As a research team, we made the conscious decision to bring as much variety into our team as possible. The team consisted of two ethicists, a paediatrician, a child psychiatrist, a sociologist and a legal expert. Because of this interdisciplinary team, there was a continuous discussion concerning the analysis, the conclusions and recommendations that followed from the results of our study. Although it did imply a lengthy writing process, it also was an important step in achieving triangulation of our findings and credibility of our analysis.²⁹

By interviewing a large number of parents, (64 parents of 44 children) and physicians (34 physicians of different medical affiliations and specialisms) this study has included a wide variety of experiences. The conscious decision was made to recruit participants with different backgrounds based on diagnosis, age, cultural background, place of residence, level of

education and the hospital where care was received. Many studies in paediatric palliative care are limited by diagnosis, often with a heavy focus on oncology. Studies are often limited to conditions with a lethal diagnosis.^{15,30-50} In this study we made the decision to include parents and physicians from children with various illnesses, in various stages of their illness, receiving care from various hospitals, providing a much needed broad perspective on suffering of children with life-threatening conditions.

During the process, some conscious decisions were made to strengthen our methodology. During recruitment, the variety of our participants was repeatedly evaluated, and adjustments to the recruitment process were made when necessary. For example, when it turned out that our initial recruitment –which used terminology such as ‘palliative’ and ‘end-of-life decision-making’— created a bias towards participants that had experience with a lethal diagnosis, the decision was made to redefine the inclusion-criteria, to include all ‘children with life-threatening conditions.’ This decision allowed our research team to also gather important experiences of participants for whom the life-threatening condition of their child did not mean embracing death. Decision-making is the balance between life-limiting and life-prolonging options, between death and life. If we would have focused only on life-limiting decisions, and only on death, the study would have described a very unbalanced picture. As this father of a girl with a life-threatening neurological tumour recalled:

“I think our oncologist had asked us before for this research. She asked us if we would like to participate in a research about end-of-life decisions, or something like that. And I said: of course not, we won’t participate, because our child is not dying, we are fighting to let her live!”
(Interview F43.)⁵¹

Throughout the entire process of this study, regular meetings were held with an advisory board of parents, researchers and physicians. They gave feedback on the progress and analysis of the research project, providing a much needed outsider-perspective, and strengthening our methodological rigour. Finally, by using and adhering to standards of reporting qualitative studies,^{52,53} we strived to objectify our results as much as possible and be transparent about our subjectivity.

2b. Limitations

Despite the strengths of our study, several limitations have to be considered. The data of this study relied on the recall of participants. Participants were asked about past experiences. The reason for retrospective interviews was both practical and ethical: interviewing participants as they went through such decisions, would have been difficult to achieve, as well as putting an unreasonable burden on the participants. The recall bias was minimized by excluding cases where the child had died more than 5 years prior to the interview, but memory and processes of grief may have influenced the participants' abilities to recall of their experiences.⁵⁴ We were however struck by the ability of participants to provide very detailed descriptions of past events, including exact dates of events that happened years before, detailed descriptions of events and names. Given these lively narratives, we believe that they provide a worthy source of data.

Second, the sensitivity of the topic may have limited participants in being complete in relaying their narratives. One could question if physicians dared to reveal their own practices. Indeed, in several interviews it became clear that physicians feared repercussions, or asked for extra measures to ensure the anonymity of their data:

“A few years ago, we learned that this girl had –and maybe you should delete this, because it might compromise my anonymity— a condition called [name of a rare metabolic condition]”
(interview P02)

Precautions were taken to ensure their anonymity; all interviews were anonymized during transcription and only the interviewer and principal researcher had access to the identifiable data of the participants. Based on their detailed descriptions, and variety of quotes where insecurities and grey areas are described, we do believe that participants were honest in telling their experiences.

Finally, my background as an ethicist may have may have led to underexposing certain (medical) aspects of decision-making. During interviews, certain medical aspects of decisions were not discussed in detail due to differences in knowledge between participant and interviewer. But at other times, the position as an outsider was an asset. In sensitive topics, parents sometimes stated they had appreciated that I was not ‘one of them’ and felt free in

criticizing health care professionals. Physicians seemed to feel less judged because I was an outsider.

2c. Children as participants

This thesis does not contain experiences of children themselves. The question is whether the lack of interviews with children themselves needs to be seen as a limitation or not. When designing this study in 2015, the aim was to include children as participants as well. Initially we tried to recruit children who were suffering from life-threatening conditions, and were capable of being interviewed. Despite serious efforts we only succeeded in including one patient (11), and one brother (9) who had lost his younger brother. The realization that most children with life-threatening illnesses are physically or mentally unable to participate in qualitative interviews, led our research team to the decision to stop recruiting them.

It is a known phenomenon that children with life-threatening conditions are particularly difficult to include in research studies. One of the reasons for this is that especially in this uncertain phase, physicians and parents show gatekeeping behaviour.⁵⁵ Children are often too ill and their time too precious to use it for medical research.^{23,55,56} Several authors argue that because of this, researchers should be careful when deciding to include patients with palliative illnesses into research.^{23,56} Especially in a study exploring not only care, but also end-of-life decision-making, these are ethical reasons to be cautious in recruiting children.

There is a second reason why judge the eventual exclusion of children to be a prudent decision, which might be best illustrated by sharing some insights into the interview with a young patient. This patient, a girl, was eleven years old, and suffered from a rare metabolic condition. Her condition gave her a lowered immune system. Illnesses or infections that were relatively harmless for most of us, were potentially life-threatening for her, if she could not get to a hospital quick enough to receive treatment. Although her illness was indeed a life-threatening condition, her life was not determined by it. When I interviewed her, she struck me as a smart, carefree girl, who couldn't wait until the interview was over so she could play with her friend again. She lived a relatively normal life with school, sports and future-plans in it. Although she was aware of her illness and the potential seriousness of it, it did not have a large role in her life. Dying was mostly a theoretical concept for her, not something she linked to herself, even though she admitted knowing that in certain specific circumstances, her condition

could become lethal. In other words: while I had expected to interview a patient, I had in fact a conversation with a child.

Child: If I would get sick, I.. Well I could die if they wouldn't help me in time or something like that.

Interviewer: Okay

Child: Yes [laughs]

Interviewer: Is that a frightening idea for you?

Child: Well, I'm not going to those countries. Usually we just stay close to the Netherlands.

In some ways this girl was a lot like the other children in the study, who despite the seriousness of their condition, remained above all: children. This focus on childhood identity is discussed in chapter five and seven. However, there was an important difference between this girl and many other children who face life-threatening conditions: she, a smart young girl, lived a life comparable to that of many healthy children. For many children with life-threatening conditions, this is not the case. Many children suffer from minor to severe physical and mental disabilities, especially since a major part of cases in paediatric palliative care concern with neurological and metabolic conditions.^{57,58} With the currently used methods of qualitative data recruitment, many of these children will never meet the criteria to be included in research: they are not communicative enough, or are physically or mentally unable to undergo the interview. The interviews with the patient and the brother did however bring us other insights, and also showed how some children talk about illness. While this may not have been our research question, the insights are no less inspiring and intriguing, as this excerpt of the interview with a brother (9 years old) of a deceased young boy shows.

Interviewer: Can you tell me a bit more about the illness of [your brother]?

Brother: Well, a brain condition is a bit like an electrical failure. You've got all kinds of little pins in your head, and they control, or operate you. But sometimes -how do you call it- they had an electrical failure, and that is why things didn't go well inside his head.

Interviewer: Okay, I understand.

Brother: And then he would get... well, it is a bit like when you're building a tall Lego Tower, but the bottom block is too small, so the tower collapses really quickly, it was a bit like that.

Whether or not a bias in the population when including children as participants is problematic, depends entirely on the research questions, but unless researchers come up with a methodology that can include patient with different mental and physical abilities, qualitative research that includes children will always have a bias.

Despite these considerations, I would argue that researchers have a duty to keep exploring ways to make the voices of children heard, even if including them as participants into traditional (qualitative) research is unfeasible and -to an extent- unethical. In future research, we can explore the possibilities of lowering the burden for children to participate in research, for example by designing methods of collecting qualitative data through different sources, such as electronic diaries to reduce physical and time constraints, or by working on methods that makes it possible for a group of patients with more limited mental or communicative capabilities express their opinion in a scientific study.

3. Answering the research questions

In this research project, we gathered and analysed the narratives of parents and physicians in order to answer our five main questions. This paragraph outlines the conclusions to all these research questions.

1. What is the current context of regulations on care and decision-making in children with life-threatening conditions?

When a young child faces a life-threatening condition, physicians and parents face a range of treatment decisions. In most cases, care will be curative or life-prolonging.^{49,59} Curative or life-prolonging care can be combined with care that aims at relieving suffering and improving quality of life: palliative care.¹ Over the last few years, Dutch paediatric palliative care has made significant steps, including the instalment of paediatric palliative care teams in all academic hospitals,⁶⁰ and the 2015 national guideline on palliative care for children.⁶¹ However, in some cases curative or palliative treatment is seen as unsuccessful in relieving a child's suffering. In such cases the decision can be made to forego life-lengthening treatment or make decisions that relieve a child's suffering but shorten a child's life-expectancy: end-of-life decisions.

Active life-ending is not regulated for children in the age between one and twelve. There are, however, other end-of-life decisions that can be made, such as withholding or withdrawing treatment, or palliative sedation. In Dutch paediatric palliative care the process between care and end-of-life decision-making can be seen as one single and continuing process. The ultimate aim of paediatric palliative care is to relieve suffering. Unique for the Dutch context, this that it explicitly translates this aim to quality of living *and* quality of dying.⁶¹ There is a strong focus on proportionality of life-prolonging treatment: every decision to provide life-lengthening is to be preceded by the deliberation whether it is in the child's best interest to do so. End-of-life decision-making and palliative care are therefore not alternatives, but parts of the same process. (Chapter 3)

Within this context, the debate on regulating active life-ending in children between the age of one and twelve needs to be understood. Currently, the Netherlands has regulations on euthanasia on request of the patient under strict criteria of due care in patient of twelve years and older,⁶² and provides a legal guideline on active life-ending in infants below the age of one.⁶³ This leaves a legal gap on regulations of active life-ending in children between the age of one and twelve. Over the last years, several voices have stated that this gap is unfair. They claimed that if we accept that can be an ethical ground to perform active life-ending, it would be unfair to deny this option for children between the age of one and twelve who could suffer unbearably as well. In this discussion it became apparent that so far, very little is known about children with life-threatening conditions in this age group. (Chapter 2)

However, regulations are not neutral: The case study of a debate between parents and physicians on providing life-prolonging treatment on a new-born girl with trisomy 18 showed that regulations shape the way physicians think about life and death. In this case study, the fact that trisomy 18 is mentioned in current regulations on active-life ending as a condition that may legitimate active life-ending because of its 'incompatibility with life', shaped the perception of the physicians. It made it difficult for them to see the child as a child that could possibly experience quality of life, and might deserve non-invasive, life-prolonging treatment. The case gives insight in the correlation between regulation and our ideas on lethality, suffering and quality of life. Regulations are not a-moral: they do not only provide freedom to make certain

decisions, but also influence the decisions we make. This calls for an open discussion on debating life-prolonging and life-limiting decisions, involving both physicians and parents. This is only possible if we present all parties involved with the whole picture: of both life and death (Chapter 4).

2. What does ‘good care’ mean for children who face a possibility of dying?

In our interviews, we explored what parents perceived as barriers to good care. Their experiences are related to six major themes: communication, organization, decision-making, end-of-life decision-making, family-care and attention for the child as person.

Communication was a major barrier in care. Although often overlooked in medical training,^{64,65} communication is of great importance in the way patients and families perceive care, something which is possibly best signified by the fact that most complaints in paediatric care are about communication, not the administration of medical care.^{66,67} Parents experienced practical barriers in communication, such as the setting of conversations, the presence of parties during the conversation, and timing. However, many parents related that bad news conversations indicating that their child might not survive, was not always held, or held only shortly before the death of the child. This meant that parents were often unprepared for the death of their child, and were denied the chance to make fully informed and well-considered decisions about their child’s treatment. Their experiences convey an important message to physicians: talk with parents about their child’s future, especially when the prognosis is uncertain. (Chapter 6)

Other barriers that parents experienced were organizational barriers, such as bureaucratic obstacles, and a lack of continuity of care. Parents often felt unsupported in overcoming these obstacles. Third, parents wished for more involvement in decision-making, and relate that important decisions were sometimes made without their input, even though parents argued that they held important knowledge on the child’s day-to-day condition, and its quality of life.

The lack of possibilities to make end-of-life decisions for their child when the child was suffering, was also explicitly mentioned as a barrier by parents. Finally, parents felt that current care for children with life-threatening conditions focuses too much on the medical, symptomatic

side of care, and has not always enough attention for the families and children that are affected by the illness. Despite the aim of palliative care to improve quality of life, a concept that (although an golden standard definition is lacking) encompasses the interests of the person's life as a whole,⁶⁸ the child behind the illness is sometimes forgotten. Paediatric palliative care might suit the needs of parents and children better when it re-evaluates its current professional detachedness, and progresses towards a medical professionalism where not only symptoms, but also people are treated (Chapter 5).

3. What does suffering mean in children with life-threatening conditions?

Given the overall aim of paediatric palliative care –to relieve suffering and improve quality of life—¹ insight in what suffering is for children with life-threatening conditions is of great importance. However, so far, little is known about suffering as a concept in general, and even more so in paediatric palliative care.

Despite the quickly developing palliative care in the Netherlands, suffering is still frequently seen in children with life-threatening conditions. They describe experiences of suffering that fall into three main categories: physical, psychosocial and existential suffering. They also describe what separates merely experiencing symptoms from becoming actual suffering for a child: a child suffers when the symptoms threaten its identity as 'child', and the child becomes a patient rather of a person. We therefore propose an experience-based concept of suffering that concentrates in the self-identification of children: suffering is when a child stops being a child. These descriptions tie in with Cassell's definition of suffering as threats to the intactness of personhood.⁶⁹ Because of the emphasis on a child's personhood, we conclude that to fully relieve suffering, the processes of dehumanisation in medicine should be re-evaluated. Although the current growing attention for patient-centred care is an important development in paediatric palliative care,⁷⁰ a further development towards 'person-centred care; may be necessary to be able to fully address a child's suffering in all its aspects. (Chapter 7 and 8)

4. What are perceptions of parents and physicians on end-of-life decision-making?

In the palliative phase, suffering of children can raise questions on the proportionality of treatment. Parents and physicians agree that in their attempts to relieve the (unbearable) suffering of a child, sometimes the best option is to stand back, provide as much comfort as possible, and accept death. In many interviews, parents and physicians related to have made such decisions, ranging from withholding curative treatment, and palliative sedation to rare cases where the dying process was initiated by withholding artificial nutrition and hydration in an otherwise stable child (a rare practice that in the Netherlands is known as ‘versterven’).^{71,72}

Decisions that influenced life-expectancy were often made long before the terminal stage of the illness. While parents acknowledged the potential life-shortening effects of such decisions, they did not perceive them to be end-of-decisions, which signifies that the current terminology of ‘end-of-life-decisions’ does not match the way such decisions are currently made. Rather than choosing between life-prolonging or life-shortening care, parents navigated a continuous range of decisions (see figure 2).

5. Do parents and physicians perceive a need for regulations on active life-ending?

In several cases parents requested active life-ending for their child, or retrospectively expressed a wish for active ending of life in the interviews. These requests generally fell into two categories: situations of suffering through a prolonging dying-process, and situation of continues suffering and lack of quality of life without other options to end the child’s suffering.

In several interviews, participants described the prolonged process of dying in children as a source of suffering for the child. In some cases, the process of dying lasted over two weeks, creating what parents saw as an undignified death, with suffering for the child. Symptoms such as epileptic seizures and pain were often seen in the terminal phase. Especially in children who had used sedatives during their treatment, physicians had difficulties to adequately sedate the child. Although sedation was often given during the terminal phase, many physicians admitted

to be hesitant about given the child a higher dosage, fearing to cross the line between palliative sedation and active life ending.

A case study of the parents of two sisters who both suffered from a lethal brain tumour, gives a deeper understanding of why parents may sometimes wish to request active-life-ending, even when it goes against their own heartfelt wish to keep their child with them. In this study we analysed the experiences of two parents, who, after losing their eldest daughter at the age of two to a lethal brain tumour, learn during the pregnancy that their second daughter suffers from the same condition. Having experienced the end of life of their eldest daughter, whose end of life had been accompanied by severe epileptic seizures with severe back-bending and (unconscious) screaming, they requested for active life-ending in their second daughter, to spare her the suffering of their eldest child. Such symptoms are not uncommon at terminal phase of children with brain tumours.

The case study also reveals the difficulty of making end-of-life decisions: while the symptoms caused by the mass effect of the tumour are very severe for bystanders, many - although not all— physicians agree that children are at this stage incapable of actively experiencing the symptoms themselves. There is no doubt that such a life-ending creates suffering for the parents, but there is no way of knowing for certain if the child suffers. This creates immense difficulties for deliberations on active life-ending, where the unbearable suffering of the patient is a major criterion: how can we decide on active life-ending with so much uncertainty about a child's suffering? Decision-making, whether it is about withholding or withdrawing kinds of treatment, terminal sedation or euthanasia, is not merely a case of regulations: it also requires a shared understanding of what suffering is.

Regulations on decision-making can aid physicians to not stand empty-handed in cases of severe suffering but they need to be accompanied by guidance on how to interpret the suffering of these children. A debate on legalization of active ending of life in children can only be successful when it is accompanied by conversations about a general consensus on the concept of suffering.

4. Ethical considerations considering active ending of life.

4a. Ethical legitimization of active ending of life.

The practise of active ending of life or euthanasia is highly debated. The *World Health Association* (WHO) is not outspoken on its position towards active ending of life or euthanasia,⁷³ but several other organisations have rejected the practice. In paediatric palliative care, the *International Children's Palliative Care Network* (ICPCN) has been outspoken in its critique against euthanasia in children.⁷⁴ The *World Medical Association* (WMA) has also stated that the practice of active ending of life –both in competent and incompetent patients— is in conflict with the basic ethical principles of medicine.⁷⁵

“Physicians-assisted suicide, like euthanasia, is unethical and must be condemned by the medical profession. Where the assistance of the physician is intentionally and deliberately directed at enabling an individual to end his or her own life, the physician acts unethically. However the right to decline medical treatment is a basic right of the patient and the physician does not act unethically even if respecting such a wish results in the death of the patient.”⁷⁵

Several critiques on active life-ending in children, are based on a critique of the practice of euthanasia in competent patients. In chapter 2, we asked several international authors to reflect on the question whether or not regulations on active ending of life should be extended to include children aged 1-12. In response, Kaczor argues that the practise of euthanasia is based on the false claim there are cases where ending a life is the only way to relieve unbearable suffering.⁷⁶

If we kill patients rather than relieving their pain, the practice of euthanasia undermines the practice of palliative care. Why worry about alleviating someone's pain, when we can simply kill the person?⁷⁶

Other critiques on active ending of life in incompetent patients focus on the fear of a slippery slope: a legalization might increase the normalcy of active ending of life, possibly even leading to situations where parents have to defend themselves for not choosing this option.^{76,77,78} While this is a genuine concern, the 15 years of experience with active life-ending in neonates indicate that such a slippery slope is unlikely to happen.⁷⁹

Not all critiques on active life-ending in children are based on a rejection of euthanasia in competent patients. Kon argues that the justification of the practice of euthanasia in competent patients does by necessity not legitimize active life-ending in incompetent patients, as euthanasia presupposes an accurate weighing of burdens and benefits.⁸⁰ He states that without the self-determination and the testament of a competent patient regarding the patient's unbearable suffering, weighing the benefits and burdens of a child's life is impossible.⁸⁰ Kon is indeed right that the value system underlying euthanasia does not support active life-ending in patients incapable of making a request for ending of life themselves. This argument has two components: autonomy (or self-determination) and the child's best interest, which will be subsequently discussed.

4b. Autonomy and the right of self-determination.

The right of self-determination (derived from autonomy), is one of the core values of euthanasia.⁸¹ Euthanasia on request of a patient younger than 12 might be possible (as indeed the Belgian legislation is built upon this presupposition)⁸², and research has shown children up from the age of eight years old are capable of making complex decision regarding their own treatment.⁸³ This group is not represented in this study, as our study found no evidence of children between the age of one and twelve requesting euthanasia. All expressed wished for active ending of life came from parents of children who – by their age, neurological condition or terminal stage of the illness- were incapable of expressing their own wishes in an explicit manner.

This has consequences for active ending of life as an ethical practice grounded in autonomy. Few topics have been as widely debated in political and moral philosophy as autonomy. Autonomy has a plethora of applications and definitions, but many –if not all—, share the aspect of self-governance.⁸⁴ The question is what 'self' here means. If 'self' merely refers to an individual human being, then active ending of life in children who are incapable of making autonomous decisions cannot be grounded in a right of self-determination. There might, however, be another interpretation of self, that extends autonomy from a first person-perspective to the autonomy of the family unit.⁸⁵ Instead of focusing on a child that might be

represented by its parents, we might focus on the family, acting as an autonomous unit. Indeed several authors, including as Bluebond-Langner et al. and Catlin, propose such a position with regard to decision-making in paediatric palliative care.^{86,87} Kon argues that such a view might be more consistent families' own perception of their relationships.⁸⁰ Although this position may not be 'universally accepted',⁸⁰ it is a position that in practice can often be observed in other medical decisions for children, as in medical decisions, parents are often included to represent both their own, and their child's interests. These questions provide an interesting basis for future research.

4c. The child's best interest.

Second, it needs to be determined how parents and physicians are able to determine the child's best interest. Some critiques interpret the Dutch stance towards active ending of life of incompetent patients as a rejection of the best interest of the patient (which may contrast the wishes of parents, physicians or even the public).^{76,78} The data from our study rejects this interpretation. We found no evidence that active ending of life was supported in lieu of what was considered to be in the child's best interest. Both advocates and opponents of regulations on active life-ending defended their views with appeals to the child's best interest, which often went against their own interest (the wish to keep their child with them). The core of the issue lies in the fact that there is no definitive answer to the question what the best interest of the child is. Even in cases where physicians and parents are in agreement about the child's unbearable suffering, the question remains: what is the child's best interest? Children with limited communication (which all children for whom active ending of life was considered in this study were) are a vulnerable group, because they are unable to defend their own interests.

Even a child that suffers unbearably, still has an inalienable right to life.⁸⁸ Is the child's best interest most supported by respecting the child's fundamental right to life, as described in the European Convention on Human Rights,⁸⁸ or by ending the child's suffering? As Zwiers argues in the context of active life ending in neonates, when the choice is between death and unbearable suffering, there is no lesser evil.⁸⁹

In this context, it is of importance to look at the life-expectancy of the children for whom active ending of life was considered. No requests were made for children for whom there was a chance of curation. In chapter 7, we distinguished two different groups of children for whom active life-ending had been considered: on the one hand children with a terminal condition, but a longer life-expectancy, and on the other hand children who were in the dying process. Participants were very sensitive to the child's life-expectancy as a factor in their request, and several felt that the question whether or not did at least in part depend on the life-expectancy of the child.

While a limited life-expectancy does not erase the child's right to life, it does seem to influence the balance between life and relief of suffering. Battin seems to make this distinction in her defence of paediatric euthanasia.⁷⁶ She argues that:

I believe that opponents would have to show evidence that at least one and perhaps many of the following propositions are true if they are to persuade you not to support this change in the law: (...) That "euthanasia" is the same as (wrongful) killing, and doesn't refer to helping someone who is already dying die in an easier, gentler way. (...)

So, dear Minister, please be as clear as you possibly can that you are only legalizing euthanasia in the Dutch sense. That is, you want to permit the ending of life in a way that, given the unbearably sad circumstances of a child's dying, can make that gentler, easier, and more humane for both the child and for the parents in whose arms you can help that death to occur.⁷⁶

The Dutch regulation on active life-ending in infants does not explicitly distinguish between children with or without a terminal prognosis, or the life-expectancy of the child.⁶³ Manninen has argued that the regulation is implicitly directed towards terminally ill children,⁹⁰ although other authors reject this interpretation.⁷⁸ Given both the moral significance and the significance that parents and physicians attach to the question, it stands to reason to include such deliberations in a debate on the legalization of active ending of life in children. Since active ending of life includes a weighing of suffering versus life, the life-expectancy of the child matters: it matters if the attempt to relieve unbearable suffering bereaves the child of years, months, or possibly days. It matters on an ethical level, and on an emotional level for parents and physicians, who may have to live with the decision.

4d. Towards a shared understanding of suffering.

The considerations highlight another ethical consideration: the lack of a shared understanding of suffering. In earlier debates on active ending of life in infants, this lack of a shared understanding of suffering also surfaces frequently. For example, Chervenak et al argue in their criticism of the regulation on active life-ending in infants: “An infant with spina bifida cannot suffer unbearably. Infants might be able to experience unbearable pain, but spina bifida does not cause it.”⁷⁷ Chervenak et al refer to a case where Dutch physicians judged the suffering of children with spina bifida to be so severe that active ending of life was justified. Chervenak et al object that these children didn’t suffer, because spina bifida itself doesn’t cause suffering.⁷⁷ This argument reveals a lack of shared understanding on the concept of suffering: is it physical pain, lack of quality of life, or something else? Chervenak refers to suffering as physical pain, the Dutch physicians the article refer to seem to use a broader definition. Throughout the entire debate on end-of-life decision-making the lack of a shared understanding of suffering is a recurring theme.

Lantos states that most requests for aid in dying are not primarily related to pain, but to loss of autonomy, not being a burden to others, depression, hopelessness, and dismissive attachment.^{76,91,92} Yet, he argues on that in children that none of these rationales are applicable to children. The definition of unbearable suffering that he envisions to be applicable to children seems largely to focus on pain and physical symptoms.

[U]nbearable suffering can usually be treated by high-quality palliative care. Patients who are in pain or who have unbearable suffering can be treated with steadily increasing doses of narcotics. Then, either their pain will be relieved or, in rare cases, they go on to respiratory failure and death.ⁱⁱ Treatment of the sorts of existential suffering and fears about the future that is the more common justification for assisted suicide or euthanasia in adults will require a very different sort of assessment and response. In those cases, the goal is not to relieve current suffering. It is to prevent the possibility of future suffering. Such concerns will generally not be relevant to children.⁷⁶

From the interviews, it seems interviewees don’t limit their understanding of (unbearable) suffering to physical pain. Both physicians and parents have a broad understanding of suffering

that includes physical, psychosocial and existential themes. Most importantly, their understanding of suffering was linked to the loss of identity: becoming a patient at the loss of being a child. This was not only true for their understanding of suffering in general, but also for situations where active ending of life was requested: such requests were based on both physical and existential forms suffering, such as loss of dignity, loss of quality of life and loss of humanity. Lack of agreement on suffering was more often focused on unknown parties, such as the unknown legislator, i.e. parents and physicians might agree, but physicians feared that a legislator might interpret suffering as merely physical pain.

While there was agreement on the causes of suffering, there was a significant lack of shared understanding on the nature of suffering. Is suffering something that we actively experience, or is it another phenomenon? For Cassell suffering is ‘the state of severe distress associated with events that threaten the intactness of a person.’⁹³ Although the literature has since both criticized and refined this definition,⁹⁴⁻⁹⁷ Cassell’s definition remains the one most often used and cited. This definition alludes to suffering as a phenomenon that is actively experienced. Many parents, and several physicians stated that suffering could also occur in children who were unaware of the phenomenon themselves. They linked a child’s suffering to the child’s dignity, which was in cases threatened by the prolonged process of dying, or symptoms displayed in the dying phase, even when the child does not actively experience these symptoms. Physicians differed in their opinion whether or not this was suffering for the child, and admitted to find requests for symptom-relief or life-ending based on these symptoms to be very difficult.

The problem with conceptualizing suffering is that there is a fear of mixing two different notions: the suffering we see in children, and the suffering that legitimizes active ending of life. Are these the same? The first is a descriptive, scientific notion of suffering, and one that has been explored in this thesis. The second question is more of an ethical and political nature. With regard to the first question, we concluded that suffering in children encompasses physical, psychosocial and existential aspects, which all appeal to the child’s identity as a child. This, however, does not imply that the suffering that would be addressed in an end-of-life decision-making shares this broadness. A good example of this distinction is the current legislation on euthanasia for children in Belgium. These regulations state that (amongst other criteria of due care) euthanasia is only permissible in cases of unbearable *physical* suffering.⁸² The fact that

Belgian legislation on euthanasia in children is limited to physical suffering does not necessarily imply that their opinion is such that a children *is* as a fact of matter only capable of physical suffering, but rather that a medical decision *ought* to be limited to physical suffering. This distinction bears resemblance Hume's well-known argument concerning the is/ought dichotomy: that we cannot simply derive normative statements from descriptive facts.^{99,100}

The Belgian decision to limit euthanasia to physical suffering may have been based on considerations with regard to the certainty with which a physicians can assess physical pain, or a consideration with regard to the limits of a physician's professional duty alleviate suffering, which one could argue encompasses pain, but not necessarily psychosocial or existential suffering.

If medical and political authorities in the Netherlands are willing to consider regulations on active ending of life in children this debate should include considerations with regard to suffering. What the result of such a debate should be, I cannot answer, but I do argue that the discussion should take place.

5. Going forward

5a. from research to regulations

In 2015, when the Dutch minister requested our research group to conduct research into care and decision-making around the end of life of children (1-12), the question was twofold: to provide insights on the and to provide recommendations on care, decision-making and active life-ending for children between the age of one and twelve.

In September 2019, our research group reported the outcomes of the three studies. Based on the results from the death certificate study, qualitative study and the questionnaire, the following conclusions were drawn.

1- Based on the data from the death certificate study and the interviews, we concluded that there is no evidence that active life ending in young children (1-12y) is currently being practised.

2- In the qualitative study, physicians did describe a grey area between palliative sedation and active life ending, where the boundaries between both actions are unclear.

3- There are currently situations in young children aged one to twelve) where children are suffering unbearably. Within the current framework of possibilities, physicians are not always able to relieve this suffering.

4- A limited group of parents and physicians express an explicit wish for more legal options on active life-ending in children aged one to twelve.¹⁹

These conclusions give insight into a practice where in certain cases children suffer, sometimes, in the eyes of parents and physicians unbearably so. This sometimes leads to a situation where parents and physicians feel that the only way to relieve the suffering is by ending the child's life.

What do the results of this study mean in the light of a political debate on regulating active ending of life in children? Does it mean that a satisfactory answer can be provided to the question whether or not regulations should be expanded to include children aged one to twelve? The short answer to that question is that it doesn't.

The question whether or not active ending of life should be regulated is a political question, not a scientific one. During this research project, we as researchers had to navigate between the scientific and political aspects of this debate. When informing the Dutch minister of Healthcare, Welfare and Sport, we as a research team made the conscious decision to limit our recommendations to the data of the study, and not to take a definitive stance on the question whether or not these findings implied an amendment of regulations. The narratives provided by parents and physicians in this study provide a plural voice, and by taking a stance, that plurality might have been jeopardized.

Our research provided insight into a context of current care and decision-making, and analysed positions of physicians and parents to uncover the arguments that underly their convictions: their conceptions of suffering, life-expectancy, decision-making and good care. Despite our hesitance to take a stance, the debate on end-of-life decisions and active life-ending is a political one. That means that going forward after this study has both a scientific and a political answer.

From a political point of view, many steps can be taken to implement the results of this study. Since we reported this study to the ministry of Healthcare, Welfare and Sports in September the first steps in this direction have been taken, including debates with the minister and several political parties. In these conversations we emphasised that implementation should not only focus on regulations for end-of-life decision-making, but on improvement of care as well.

5b. Recommendations for future research

This research answers important questions in a field where so far, very few answers were available, but it also leads to new questions. The scientific development in paediatric palliative care has been remarkable. As Sisk et al describe in their historic overview, paediatric palliative care has developed in a mere matter of decades from 'veritable neglect to the development of paediatric palliative care as a subspecialty devoted to their care.'¹⁰⁰ Nevertheless, paediatric palliative care is still a recent development, and there is still much to be known about end-of-life care and decision-making in children. 5 fields for possible further research are:

1) Throughout this thesis, participants underline the importance of person-focused care: what it means to be human, and what it means to be a child. Despite its importance, parents feel that this human-focused side of care remains underexposed. Despite its totalitarian aim, guidelines are care plans paediatric palliative care remain predominantly symptom-focused.^{2,101} Future research into existentialist aspects of care for children with life-threatening conditions might help to achieve the goal of a 'total care' in paediatric palliative care.¹⁰²

2) This study also highlighted the grey area between active ending of life and palliative sedation. Physicians reported to find it difficult to the line between symptom-relief and active

ending of life. More research might contribute to further define this grey area, and provide clarity to physicians who face such decisions.

3) Third, this study was limited to children between the age of one and twelve. The results of this study are not applicable to older children, but similar themes might exist (and there are signs that they indeed do exist) in older, incompetent patients as well. Especially themes such as active ending of life in this age group have not yet been methodically been researched in this group. Future research into this age group may clarify the specific needs of such children and families.

4) A final recommendation for future research is to bridge the gap between practise and policy. In the interviews, many physicians stated they not only did they lack legal possibilities to act in cases of unbearable suffering, but they also lacked guidance on when an appeal to such a regulation would be justified, how to assess suffering and quality of life, and how to respond to requests from parents (or hypothetically: children) for active ending of life. This lack of guidance was experienced with regard to all end-of-life decisions. In chapter 9, we concluded that regulations on active ending of life need to be accompanied by a system that provides such guidance in order to be effective. In adult euthanasia (and to some extent in active ending of life in neonates) jurisprudence provides such guidance, without by creating a system of previous cases that physicians can compare to. Future research might explore options to extrapolate such a system to this age group.

Closing words.

October 15, 2020.

As I am writing the closing words to my PhD-thesis, not only a research project has reached a milestone, but a political process as well. On Tuesday 13 October 2020, one day after my supervisors gave me permission to submit my PhD-thesis, we received the news that the minister of Health, Welfare and Sport had agreed to expand regulations to allow active ending of life in children between the age of one and twelve, in cases of unbearable suffering.¹⁰³⁻¹⁰⁶

During my research project, I have often been asked by colleagues, friends and family what my own point of view on active ending of life in young children was. I have always hesitated whether or not I should answer such questions. Chapter 2, where we were asked to voice our own advice on the matter of regulating active ending of life, was therefore perhaps the most difficult chapter in this thesis. Only after long, and -admittedly- intense debates within our research team, did we agree on the phrasing that we would advise the minister to *consider* expanding regulations for children between the age of one and twelve.

I have often reflected on why stating my own position became so particularly difficult to answer, and I think the answer has to do with the way I have interpreted my role as a researcher. I have always tried to be, above all else, a listener. In order to fully embrace the narratives of others, I suspended my own opinions as much as possible. The narratives in this study were first and foremost characterized by their pluriform nature, and the data contained a wide variety of experiences and opinions. In this thesis I have tried to do justice to this pluriformity: to address both life and death, both care and end-of-life decision-making, both the suffering and the possibilities of achieving quality of life for seriously ill children.

To answer the question what my position towards active ending of life is, is that in my opinion, my own opinion should not matter: I wouldn't have been a good researcher if it did. What I *do* think, however, is that the minister's decision to expand regulations is an important step in listening to the needs of parents and professionals in the field of paediatric palliative care. And as the regulation provides merely a possibility, a regulation should not impede the wishes of families who do not wish to make such decisions.

This thesis may have reached its conclusion, but this is not the end of my ambitions as a researcher. Despite the developments, there is still a lot to achieve in pediatric palliative care. Care, communication and decision-making can, and should be further developed to suit the needs of children and families. In that development, I hope that above all, we don't lose sight of the human beings. Developing paediatric palliative care is not only about the *treatment* of *patients*, but just as much about the *care* for *human beings*.

- Marije Aafke Brouwer.

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