Palliative Care in Pediatric Oncology:

Ethical Considerations Surrounding Shared Decision-Making

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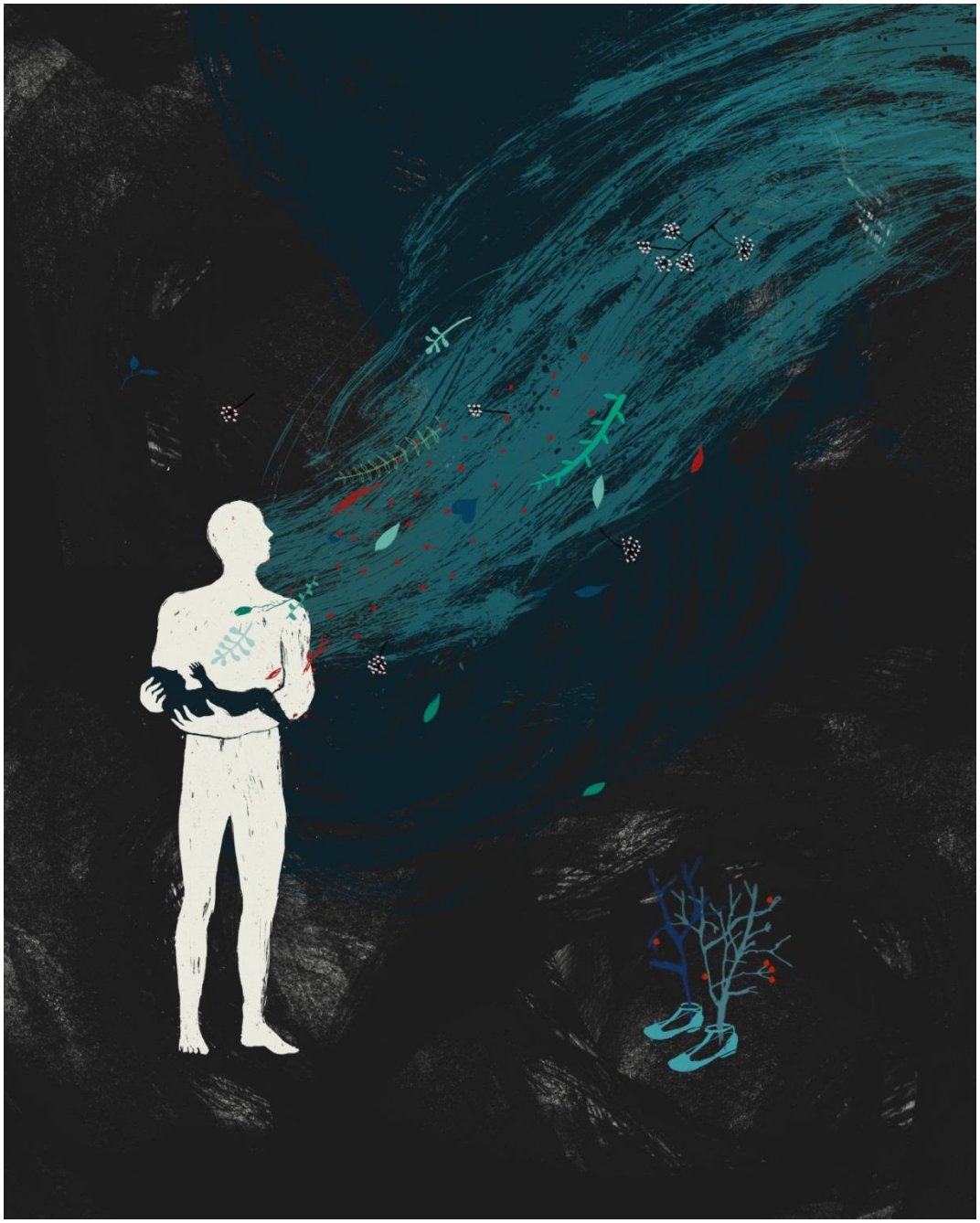

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Contributions

The study was conceived and supervised by Prof. Dr. med. Bernice Elger, head of the Institute for Biomedical Ethics at the University of Basel. She and Dr. Tenzin Wangmo wrote the proposal to receive a Grant from the Swiss National Science Foundation (Grant.-No.: 406740_139283 / 2). Prof. Dr. med. Felix Niggli and Prof. Dr. med Kühne approved the research proposal and were co-applicants. Dr. Tenzin Wangmo and Dr. Eva De Clercq drafted the study tools. I, Dr. Wangmo and Dr. De Clercq prepared the submission to obtain ethical approval for an amendment from the leading ethics committee (Ethikkommission beider Basel, approval number 51/12). I (focus groups and medical records in the German-speaking part of Switzerland), Dr. Pacurari (focus groups and medical records in the French-speaking part), Dr. De Clercq (medical records in the Italian-speaking part, focus groups in the French-speaking part), and Milenko Rakic (medical records and focus groups in the German-speaking part) carried out data collection. Further, I carried out participant recruitment (focus groups), data management and data analysis.

This thesis is arranged in three main parts: (1) background (chapter 1), (2) journal articles (chapter 2 – 9, each chapter comprising one article), and (3) general discussion (chapter 10). In the following, my contributions to each journal article as well as the contributions of the co-authors are presented in detail. The articles appear in the same order as in the thesis.

Rost M, Wangmo T, Niggli F, Hartmann K, Hengartner H, Ansari M, Rischewski J, Beck-Popovic M, Kühne T., Elger B. Parents' and Physicians' Perceptions of Children's Participation in Decision-making in Paediatric Oncology: A Quantitative Study. *Journal of Bioethical Inquiry*. 2017;14(4):555-565.

I carried out the data management, data analysis, and drafted the initial manuscript. Tenzin Wangmo supervised data collection and data analysis, and supported with the manuscript preparation. Felix Niggli, Karin Hartmann, Heinz Hengartner, Marc Ansari, Pierluigi Brazzola, Johannes Rischewski, Maja Beck-Popovic, and Thomas Kühne supported the construction of the study questionnaire and data collection. All authors critically reviewed the manuscript.

Rost M, Acheson E, Kühne T, Ansari M, Pacurari N, Brazzola P, Niggli F, Elger B, Wangmo T. Palliative Care in Swiss Pediatric Oncology Settings: A Retrospective Analysis of Medical Records. *Journal of Supportive Care in Cancer*. 2018;26(8):2707-2715.

Elaine Acheson and Michael Rost contributed equally to the writing of this paper and, therefore, share first authorship. I carried out data collection, data management, and quantitative data analysis. Elaine Acheson carried out the data entry and qualitative data analysis. Nadia Pacurari contributed towards data collection. Felix Niggli, Marc Ansari, Pierluigi Brazzola, and Thomas Kühne facilitated data collection. Tenzin Wangmo and Bernice Elger supervised data collection and data analysis. They supported with the manuscript preparation. All authors critically revised the initial manuscript for content and interpretation of the data.

Rost M, Wangmo T, Rakic M, Acheson E, Rischewski J, Hengartner H, Kühne T, Elger B. Burden of Treatment in the Face of Childhood Cancer: A Quantitative Study Using Medical Records of Deceased Children. *European Journal of Cancer Care*. 2018;27(6):e12879.

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De Clercq E, **Rost M**, Rakic M, Wangmo T, Elger B. The Conceptual Understanding of Pediatric Palliative Care: a Swiss Healthcare Perspective? BMC *Palliative Care.* 2019.

I carried out data collection, data management, and supported with qualitative data analysis. Eva De Clercq took the lead in writing the manuscript, and carried out data collection, data management and qualitative data analysis. Milenko Rakic supported with data collection, data management, and data analysis. Tenzin Wangmo and Bernice Elger supervised data collection and data analysis. They supported with the manuscript preparation. All authors critically revised the initial manuscript for content and interpretation of the data.

Cheng B, **Rost M**, De Clercq E, Elger B, Wangmo T. Palliative Care Initiation in Pediatric Oncology Patients: A Systematic Review. *Cancer Medicine*. 2018.

I contributed to study design, data collection, analysis and interpretation, and provided critical revisions to the manuscript. Brian Cheng contributed to study design, data collection, analysis and interpretation, and drafted the initial manuscript. Eva De Clercq contributed to analysis and interpretation, and provided critical revisions of the manuscript. Bernice Elger contributed to study design and critically revised the manuscript. Tenzin Wangmo contributed to study design, data collection, analysis and interpretation, and critically revised the manuscript. All authors approved the final version of the manuscript.

De Clercq E, **Rost M**, Pacurari N, Elger B, Wangmo T. Aligning Guidelines and Medical Practice: Literature Review on Pediatric Palliative Care Guidelines. *Palliative and Supportive Care*. 2017;15(4):474-489.

I carried out the systematic literature search and contributed towards refining the original manuscript draft by critically revising it. Eva De Clercq took the lead in writing the manuscript, supported the literature search, read abstracts and full texts to screen the records for eligible documents, and did the data analysis. Nadia Pacurari contributed to screening the titles and

abstracts. Tenzin Wangmo supported the literature search. Tenzin Wangmo and Bernice Elger supported with manuscript preparation, All authors critically reviewed the manuscript.

Rost M, De Clercq E, Wangmo T, Elger B. The Need for a Shared Understanding: Domains of Care and Composition of Team in Pediatric Palliative Care Guidelines. *Journal of Hospice & Palliative Nursing*. 2017;19(6):556-564.

I carried out the systematic literature search as well as qualitative data analysis. Furthermore, I drafted the initial manuscript. Eva De Clercq supported the literature search, screened the titles and abstracts for eligible documents, and supervised data analysis as well as manuscript preparation. Tenzin Wangmo supported the literature search. Tenzin Wangmo and Bernice Elger supported with manuscript preparation. All authors critically reviewed the manuscript.

De Clercq E, Ruhe K, **Rost M**, Elger B. Is Decision-Making Capacity an "Essentially Contested" Concept in Pediatrics? *Medicine, Health Care and Philosophy*. 201720(3):425-433.

I read and critically revised the whole manuscript. I also provided permission for submission and publication. Eva De Clercq took the lead in writing the manuscript. She developed the idea for this theoretical article based on her KätheZingg grant of the Swiss Academy of Medical Sciences. Dr. Ruhe provided substantial contributions to the interpretation of the theoretical background and to the focus of the article. Prof. Elger critically reviewed the manuscript.

Summary

In Switzerland, between 2005 and 2014 there was an average of nearly 250 new cases of cancer in children per year. Based on a five year survival rate of more than 80%, almost 50 deaths due

to childhood cancer per year can be estimated. According to the World Health Organization and international medical guidelines, pediatric palliative care should begin at the diagnosis of a life-threatening illness, continue throughout a child's illness trajectory, through death and beyond. In the pediatric setting, shared decision-making has become the predominant approach to facilitate the decision-making process among physicians, parents, and the child. However, there is evidence that shared decision-making in pediatric oncology needs further improvement and that it is still understudied. Thus, the goal of this thesis is to propose ethically sound and practically feasible ways to enhance shared decision-making regarding palliative care in pediatric oncology.

In order to meet this objective, a mixed-methods approach that employed various quantitative and qualitative methods is used. Furthermore, this thesis integrates empirical and normative analyses for addressing ethical concerns that contribute to the overall bioethical debate on shared decision-making regarding palliative care in pediatric oncology. The empirical bioethics methodology used to integrate empirical and normative analyses is Critical Applied Ethics. The thesis is arranged in three main parts: (1) background (chapter 1), (2) journal articles (chapter 2 – 9), and (3) general discussion (chapter 10).

First, the *background* (chapter 1) provides an introduction to pediatric palliative care, to shared decision-making, and to the ethico-legal imperative for shared decision-making. Further, after a general overview over the emerging field of empirical bioethics it delineates the particular empirical bioethics methodology that was used in this thesis, namely Critical Applied Ethics. Finally, the background presents the research objectives and the description of the mixed methods approach.

Second, each of the following eight chapters (chapter 2 – 9) comprises one journal article. *Chapter 2* gives insights into how shared decision-making occurs in Swiss Pediatric Oncology

Group centers from the viewpoints of parents and physicians. Quantitative analysis of questionnaires reveals systematic differences between parents' and physicians' perceptions of the same child. Further, sex and age predict a child's involvement in decision-making.

Chapter 3 explores the provision of pediatric palliative care and related decision-making in Swiss pediatric oncology settings. Results from a retrospective review of medical records of deceased children show that there are still high numbers of late or non-referrals and even children older than 12 years were not involved. Also, leukemia patients were less likely to receive palliative care than the overall sample.

Chapter 4 examines children's and their families' burden due to adherence to cancer treatment. Quantitative analysis of medical records of deceased children reveals that children and their families face a significant burden of treatment. Moreover, leukemia patients had a higher number of inpatient stays, spent more time in hospital both during the entire illness and during the last month of their life, and were more likely to die in the hospital than patients with CNS neoplasms and other diagnoses.

Chapter 5 addresses the understanding of pediatric palliative care in pediatric oncology centers in Switzerland. Qualitative analysis of focus group interviews with various stakeholders show that there are still difficulties regarding the implementation of the rather philosophical definition of pediatric palliative care as set out by the World Health Organization and that palliative care, frequently, is not initiated at diagnosis.

Chapter 6 evaluates the timing of palliative care in pediatric oncology patients through a systematic literature review. Data reveals that pediatric palliative care consultation does not occur until late in the illness and palliative care does not begin until close to death.

Chapter 7 identifies barriers and recommendations for the proper implementation of pediatric palliative care through a systematic literature review of articles that discuss international guidelines. Identified barriers in the literature are: gaps within medical practice, lack of evidence-based research, absence of clear guidance regarding bereavement care, and challenges involved in multidisciplinary teams. Common recommendations that can be found in the literature are: training and education, multidisciplinarity, research on the benefits of and raising awareness about pediatric palliative care. Finally, the question arises whether pediatric palliative care has not lost sight of end-of-life issues, focusing too narrowly on quality of life.

Chapter 8 covers conceptual confusion of three core domains of pediatric palliative care. Five online databases were searched systematically, in addition to a google search. Analysis focused on the language used to determine the domains of pediatric palliative care. Analysis revealed that, first, psychological care is not sufficiently demarcated from emotional care; second, it remains unclear what separates social from psychosocial care; third, spiritual care is not sufficiently distinguished from existential and religious care. Finally, it is shown that this confusion negatively affects clinical practice.

Chapter 9 argues that decision-making capacity would benefit from being treated as an essentially contested concept as this warns against any reification of what it means to have capacity. Further, using decisional capacity in a questioning mode gives space to alternative interpretations that might advance the ongoing debate surrounding decision-making.

Third, the *general discussion* (chapter 10) provides an integration of empirical results and ethical analysis which is structured in three subparts: conceptual background of shared decision-making, pre-existing conditions of shared decision-making, and the process of shared decision-making. Moreover, it illustrates the application of the used empirical bioethics methodology (Critical Applied Ethics) within the reported research project. Next, limitations

and implications for further research are addressed. The thesis closes with the conclusions section that contains seven recommendations for practice and theory which represent the very essence of the reported findings.

Zusammenfassung

Zwischen 2005 und 2014 erkrankten in der Schweiz jährlich etwa 250 Kinder an Krebs. Dies bedeutet bei einer 5-Jahres-Überlebensrate von knapp mehr als 80%, dass schweizweit durchschnittlich etwa 50 Kinder pro Jahr aufgrund ihrer Krebserkrankung sterben. Gemäss Weltgesundheitsorganisation und internationaler medizinischer Richtlinien sollte Pediatric Palliative Care bei der Diagnosestellung einer lebensbedrohlichen Erkrankung beginnen und für die Dauer der Erkrankung sowie über den Tod des Kindes hinaus zum Tragen kommen. Im pädiatrischen Setting hat sich die partizipative Entscheidungsfindung als präferierter Weg, eine von Ärzten, Eltern und dem Kind gemeinsam gefundene Entscheidung zu treffen, durchgesetzt. Allerdings zeigt eine Vielzahl von Studien, dass die Umsetzung der partizipativen Entscheidungsfindung in der Praxis nach wie vor verbessert werden muss sowie dass es weiterer Forschung zum Thema bedarf. Vor diesem Hintergrund ist es Ziel dieser Arbeit, ethisch vertretbare sowie praktisch umsetzbare Möglichkeiten aufzuzeigen, die partizipative Entscheidungsfindung bezüglich Palliative Care in der pädiatrischen Onkologie weiter zu verbessern.

Um dieses Ziel zu erreichen wurde ein Mixed-Methods-Ansatz, bestehend aus verschiedenen qualitativen und quantitativen Methoden, gewählt. Zudem integriert die vorliegende Arbeit empirische Forschung und normative Analyse, um ethisch relevante Aspekte zu adressieren und um somit schliesslich die bioethische Debatte zu partizipativer Entscheidungsfindung bezüglich Palliative Care in der pädiatrischen Onkologie zu bereichern. Die genutzte empirische Bioethik-Methodologie ist Critical Applied Ethics. Die vorliegende Arbeit ist in drei Hauptteile gegliedert: (1) Hintergrund (Kapitel 1), (2) Artikel in Fachzeitschriften (Kapitel 2 – 9) und (3) Diskussion (Kapitel 10).

Kapitel 1. Der Hintergrund führt in die Bereiche Pediatric Palliative Care, partizipative Entscheidungsfindung und in den ethisch-rechtlichen Rahmen ein. Darüber hinaus wird, nach einem Überblick über das Feld der empirischen Bioethik, die in der vorliegenden Arbeit gewählte empirische Bioethik-Methodologie beschrieben (Critical Applied Ethics). Schliesslich umfasst der Hintergrund die Forschungsziele sowie Ausführungen zum gewählten Mixed-Methods-Ansatz. Jedes der folgenden acht Kapitel (Kapitel 2 – 9) besteht aus einem Fachzeitschriften-Artikel.

Kapitel 2 liefert Einblicke in die partizipative Entscheidungsfindung in Spitälern der Schweizerischen Pädiatrischen Onkologie Gruppe. Die quantitative Analyse der von Eltern und Ärzten ausgefüllten Fragebögen zeigt systematische Unterschiede zwischen der elterlichen und ärztlichen Perspektive auf das Kind. Ausserdem sagen das Alter und das Geschlecht eines Kindes dessen Involvierung in die Entscheidungsfindung vorher.

Kapitel 3 befasst sich mit der Durchführung der Pediatric Palliative Care auf Kinderkrebsstationen in der Schweiz. Die Ergebnisse der retrospektiven Auswertung der Spitalakten an Krebs verstorbener Kinder verdeutlichen, dass nach wie vor viele späte Überweisungen in die Palliative Care stattfinden, mitunter sogar gar keine. Auch werden manche Kinder, die älter als 12 Jahre sind, nicht in die Entscheidungsfindung einbezogen. Darüber hinaus erhalten an Leukämie erkrankte Kinder seltener Palliative Care.

Kapitel 4 untersucht die Belastung infolge der Krebsbehandlung auf Seiten des Kindes und der Eltern. Die quantitative Analyse der Spitalakten der verstorbenen Kinder zeigt, dass Kinder und ihre Familien infolge der Krebserkrankung hoch belastet sind. Zudem sind Leukämiepatienten häufiger und länger (über die gesamte Erkrankung hinweg sowie im letzten Monats ihres Lebens) in stationärer Behandlung, und sterben häufiger im Spital als Kinder mit anderen Diagnosen.

Kapitel 5 befasst sich mit dem Verständnis von Pediatric Palliative Care innerhalb der pädiatrischen Onkologie in der Schweiz. Die qualitative Analyse der Fokusgruppen-Interviews mit verschiedenen Stakeholdern macht deutlich, dass Schwierigkeiten bei der Implementierung der eher philosophischen Weltgesundheitsorganisations-Definition der Pediatric Palliative Care bestehen. Auch wird Pediatric Palliative Care häufig nicht zur Diagnose begonnen.

Kapitel 6 untersucht mithilfe eines systematischen Literatur-Reviews den Zeitpunkt der Initiierung der Palliative Care in der pädiatrischen Onkologie. Die Daten zeigen, dass die erste Konsultation zu Pediatric Palliative Care spät im Krankheitsverlauf initiiert und dass Pediatric Palliative Care selbst erst kurz vor dem Tod begonnen wird.

Kapitel 7 identifiziert vermittels eines systematischen Literatur-Reviews von Artikeln, die internationale Richtlinien diskutieren, Barrieren und Empfehlungen zu einer angemessenen Implementierung der Pediatric Palliative Care. Identifizierte Barrieren sind: Schwierigkeiten in der medizinischen Praxis, Mangel an evidenz-basierter Forschung, Abwesenheit klarer Richtlinien bezüglich Trauerpflege und Herausforderungen durch multidisziplinäre Teams. Häufige Empfehlungen, die in der Literatur gefunden wurden, sind: Training und Ausbildung, multidisziplinäre Teams, Forschung zum Benefit der Pediatric Palliative Care sowie zur Sensibilisierung der Bevölkerung für diese. Schliesslich stellt sich die Frage, ob Pediatric Palliative Care sich nicht zu weit von ihrem ursprünglichen Fokus auf End-of-Life-Care entfernt hat und sich nunmehr zu sehr auf das Thema Lebensqualität konzentriert.

Kapitel 8 analysiert die Konzeptualisierung der Domänen der Pediatric Palliative Care. Hierzu wurden fünf online Datenbanken durchsucht sowie eine Google-Suche durchgeführt. Die Analyse fokussierte auf die Sprache, die genutzt wurde, um die Domänen der Pediatric Palliative Care zu beschreiben. Es zeigt sich, dass, erstens, der Begriff „psychological care“ nicht hinreichend gut vom Begriff „emotional care“ abgegrenzt wird; dass es, zweitens, unklar

ist, was die Begriffe „psychosocial care“ und „social care“ unterscheidet; sowie, drittens, dass der Begriff „spiritual care“ nicht klar genug von „existential care“ und „religious care“ unterschieden wird. Schliesslich wird aufgezeigt, wie diese unzureichenden Konzeptualisierungen die klinische Praxis negativ beeinflussen können.

Kapitel 9 postuliert, dass es vorteilhaft ist, das Konzept „decision-making capacity“ als ein „essentially contested concept“ zu verstehen. Zudem warnt es davor, die Frage danach, was es heisst „decision-making capacity“ zu haben, schlichtweg mit einer Vergegenständlichung des Konzepts zu beantworten. Abschliessend bietet diese Sichtweise die Möglichkeit, alternative Ansätze, welche die Debatte voranbringen können, ernsthaft zu erwägen.

Kapitel 10 stellt eine umfassende Diskussion der vorher aufgeführten Forschungsergebnisse dar. Dazu werden empirische Ergebnisse und ethische Analyse zusammengebracht und in drei Abschnitten mit jeweils verschiedenem Schwerpunkt präsentiert: konzeptueller Hintergrund partizipativer Entscheidungsfindung, der partizipativen Entscheidungsfindung vorausliegende Bedingungen sowie der Prozess partizipativer Entscheidungsfindung selbst. Weiterhin wird dargestellt, wie die empirische Bioethik-Methodologie (Critical Applied Ethics) innerhalb der vorgestellten Forschung angewandt wurde, bevor die Limitierungen und Implikationen für zukünftige Forschung diskutiert werden. Den Abschluss der vorliegenden Arbeit bilden sieben Empfehlungen für Praxis und Theorie, welche auf den vorgestellten Ergebnissen aufbauen und gewissermassen deren Kerngehalt widerspiegeln.

Chapter 1 – Background

1 Introduction

1.1 Pediatric Palliative Care

Pediatric palliative care (PPC) is an active and holistic approach to care for children with life-limiting and life-threatening illnesses which embraces multiple domains of care, such as psychological, physical, social, and spiritual care[1-3]. Due to its multifaceted nature, PPC requires that numerous factors are fulfilled to allow children[1] and their families to live to their fullest potential despite the life-threatening condition they are facing. In clinical practice, the absence of a single factor (e.g. a required skill or service, involvement of family in decision-making), already compromises the provision of PPC. Such an understanding borrows from a notion of Aristotle, who, in the Nicomachean Ethics, demarcated erroneous and correct actions by means of distinguishing between polymorphism and monomorphism: "Further, it is possible to err in many ways (…), whereas there is only one way to be correct. That is why erring is easy and being correct difficult, since it is easy to miss the target but difficult to hit it."[4, NE, II.6, 1106a29-32, trans. Reeve]

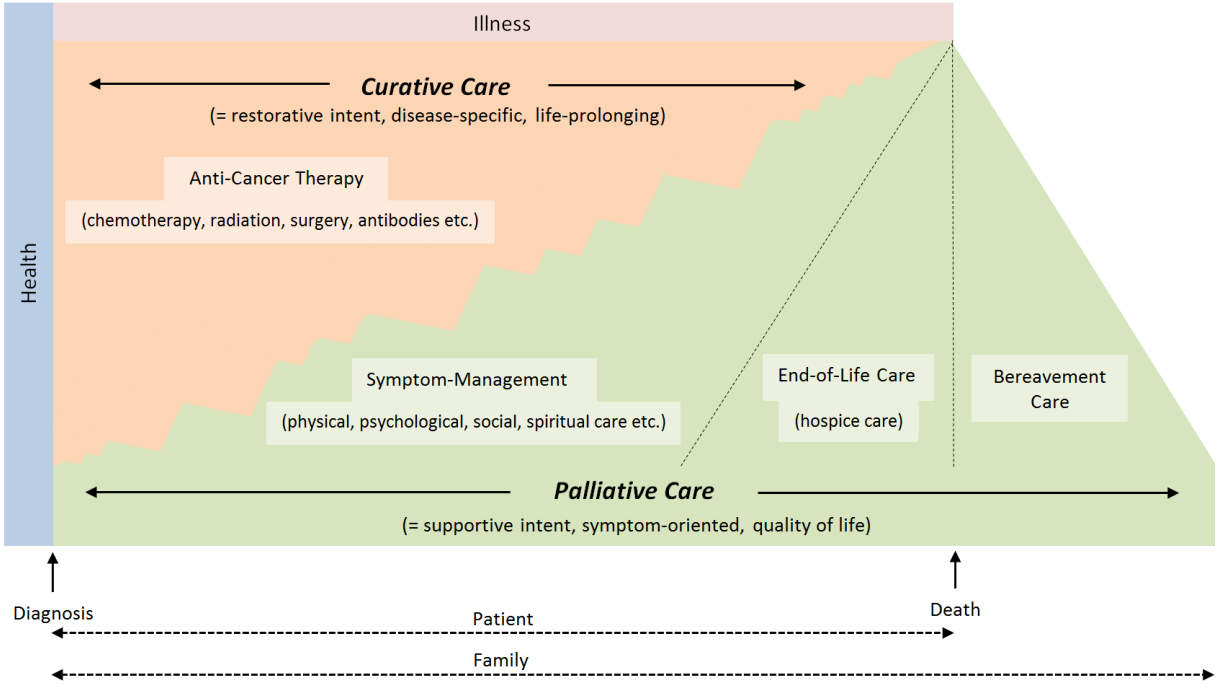
Per analogiam, adequate PPC provisions are all alike with respect to one critical feature. In all those cases the needs of the child and the family are met (monomorphic: the "one way to be correct"). In contrast, inadequate PPC provisions can take different forms because at least one of the various needs is insufficiently met (polymorphic: the possibility "to err in many ways"). Hence, the Aristotelean view offers a useful perspective on the provision of PPC: there is one ideal way of providing PPC, namely tailoring care to the particular needs of the child and the family, and there are numerous ways of providing inadequate PPC, namely not meeting or mistakenly addressing one or more of these needs. Further, this perspective implicitly

[1] The terms "child", "adolescent", and "minor patient" are used to denominate persons who have not yet attained the legal age of majority. Subsequently, if not stated differently, the term "children" collectively refers to infants, children, and adolescents.

disqualifies the notion of a universal way to provide PPC that can be applied across cases and situations. Instead, it affirms the idea of some sort of clinical casuistry, according to which PPC has to consider the particular needs of the child and the family, while taking into consideration relevant circumstances. Thus, within each palliative situation provided care has to correspond to the individual patient and its family and if so, it is adequate. The purpose of this thesis is to cast light on some of the reasons for not providing adequate PPC.

As lined out by the World Health Organization and emphasized by international guidelines, PPC begins at the time of diagnosis and continues, irrespective of whether treatment is also directed at curing, throughout a child's illness-trajectory, through death and beyond (figure 1)[1,3,5,6]. Furthermore, it is provided by a multidisciplinary team and focuses on the enhancement of quality of life of the child and his or her family[1]. Under all circumstances, PPC must be an individually tailored endeavor that is strongly oriented towards the idiosyncratic needs of the child and the family[3]. This life-affirming approach is further characterized by a general emphasis on communication through the application of shared decision-making (SDM)[7]. Finally, it is part of the philosophy of PPC that every child with a life-limiting or life-threatening condition must have access to it[3]. According to the Association of Children's Palliative Care, four categories of life-limiting and life-threatening conditions can be distinguished based on the properties of the illness, such as diagnosis and severity[3]. Pediatric cancer patients are assigned to category 1, which contains life-threatening conditions that are theoretically curable. For these children, PPC services must be accessible when the condition deteriorates acutely or in cases of failed curative measures, become dispensable in cases of long term remission or cure, and need to be assessed at any point along the illness trajectory for each child individually[3]. One important tenet of PPC is that this assessment has to be based on a child's needs but not on diagnosis or prognosis per se.

Figure 1. Model of pediatric palliative care



Note. Adapted from World Health Organization[8].

Despite improved 5-year-survival rates of more than 80% in high-income countries, cancer is still the leading disease-related cause of death in children in the western world[9]. Worldwide, annual incidence rates of childhood cancer vary between 50 and 200 per million (overall 140) for children aged between 0 and 14 and between 90 and 300 per million (overall 185) for adolescents aged between 15 and 18[10,11]. Moreover, empirical data shows a global increase of 13% in incidence rates in 2001-2010 as compared to the 1980s[10]. For Switzerland, between 2005 and 2014, cancer was diagnosed in 1.987 children aged 0 to 14 (161 per million) and in 501 adolescents aged 15 to 20 (168 per million), corresponding to 249 childhood cancer diagnoses per year[9]. Accordingly, based on a 5-year-survival rate of more than 80%, nearly 50 annual deaths due to childhood cancer can be estimated for Switzerland.

Although palliative care was developed four decades ago[12], recent studies from various countries indicate that parts of the public are still unfamiliar with this type of care[13-16]. Also within clinical practice, it is still often confused with hospice care and end-of-life care. Those

practitioners who were able to demarcate the concepts commonly associated palliative care with death and dying, and, moreover, palliative care has been fraught with uncertainties about how to put it into practice[13,17,18]. Hence, conceptual clarification of the concept is a prerequisite for research in the field of PPC. The distinction between palliative and hospice care traces back to 1990, when the World Health Organization formally delineated palliative and hospice care for the first time[19]. In contrast to hospice care that was closely related to end-of-life care, palliative care's earlier applicability in the illness trajectory was underlined[8]. In 2002, the World Health Organization articulated a revised definition of palliative care, which adds that palliative care ought to be provided for any person with a life-threatening illness[6]. Delving into the characteristics of hospice care, a conceptual analysis found that, compared to palliative care, hospice care is more narrowly defined as a community based program for patients with terminal illnesses (frequently equated with expected survival of less than 6 months), in which volunteers play a major role and which focuses on bereavement care[20]. Furthermore, hospice care does not include curative attempts, implicitly acknowledging that death is inevitable, and is provided in an inpatient-setting[21]. Hospice care can be considered as a particular model for the delivery of palliative care at the end-of-life aiming to improve the quality of death and dying[21].

1.2 Theoretical Background: Shared Decision-Making

Decision-making regarding the initiation of palliative care is burdensome and fraught with uncertainties that cause high levels of psychological strain on the part of the parents[22]. Moreover, numerous aspects have to be considered, such as: age of the child, capacity to make treatment decisions, disease experience, and finally, how, when, and by whom a decision should be made. Different types of decisions in the pediatric setting have evoked different legal

and ethical[2] debates: decisions about who decides (e.g. parents versus competent adolescents), decisions about treatments (e.g. stopping or not starting treatments, life-sustaining treatments), and decisions about the criteria for decisions (e.g. quality of life or resources)[23]. Against the background of medical ethics' transition from paternalism towards respecting individual autonomy[24], SDM has become the predominant approach to pediatric medical decision-making, especially for children suffering from chronic health conditions[25].

Generally, decision-making in pediatrics is characterized by including multiple steps and involves at least three parties: the health care provider (e.g. physician, nurses), parents[3], and patient. This triad shares the process aiming to arrive at a decision that is in the best interest of the child. However, this triadic decision-making represents a major challenge in this context[26]. Literature stresses four main attributes of SDM: (1) at least two parties are involved, (2) the parties share all relevant information, (3) parties actively work towards a consensus, and (4) an agreement is reached on the course of treatment[27,28]. Besides, informing and involving the child in a developmentally appropriate way is universally recommended by international medical guidelines on decision-making within PPC[1-3,7,29-34].

With respect to PPC, decision-making is structurally equal (triadic constellation), but it appears to be in particular emotionally burdensome and difficult for all involved parties[22]. Decisions on discontinuing curative treatment in cases of progression, not starting curative treatment in cases of relapse, or not resuscitating the child in cases of a life-threatening emergency represent important challenges.

[2] The terms "ethics" and "morality" or "ethical" and "moral", respectively, are often used interchangeably. Throughout this thesis, both terms are linguistically distinguished. Whereas "ethics" refers to a philosophical discipline that systematically and methodology-based reflects upon morality, the term "morality" refers to societal and personal value-systems that are embedded in the lifeworld. However, when referring to approaches by other authors, their terminology is maintained and, therefore, may be different.

[3] Throughout this thesis, the term "parents" does not only refer to biological parents, but to caretakers of a child in general. Hence, it also includes, for example, stepparents who adopted their child(ren) or foster parents.

In their "decisional priority in pediatric oncology model", Whitney and colleagues present a decision-making model for treatment decisions that starts from the distinction between decisional authority and decisional priority[35]. The former refers to the nondelegable right of the parents to decide, while the latter refers to identifying the single best course of action. Further, they delineate two pivotal attributes of decisions for pediatric cancer patients, namely whether the illness is curable and whether there is a superior treatment option. These two continuous dimensions represent two axes of a decisional plane in which all pediatric oncology decisions can be located. In situations where there is little chance of a cure (unlikely curability) and exclusive palliative care services are the best possible treatment (one best option), this model proposes that the physician takes the lead. Exercising decisional priority, the physician should explain to the parents (and child) that there are no curative options left and that, henceforth, symptom management has to be prioritized over continued curative therapy. Of course, decisional authority remains with the parents who are legally and ethically responsible to promote their child's interests. However, as the child matures, decisions are increasingly shared with the child. Apart from this model that understands itself as both descriptive and normative[35], several international and national, ethical and legal documents are relevant for SDM in the context of PPC (figure 2).

1.3 The Ethico-legal Imperative for Shared Decision-Making

Since direct inferences from descriptive data to normative claims are invalid, a normative framework is required to evaluate empirical data's significance for normative conclusions[36]. The ethico-legal framework presented in this chapter meets general quality criteria for a normative background, namely, coherence, simplicity, and clarity. At the same time, it meets more specific quality criteria for the selection of an ethical theory in empirical ethics, as evolved by Salloch and colleagues who plead for a systematic and reasoned selection of ethical

theories[36]. First, the ethico-legal framework is adequate for decision-making regarding PPC, since it is comprised of universally accepted principles and rights, specific guidelines on PPC, and relevant legislation. Second, it is suitable for the purposes and design of this thesis, as the purpose is to examine clinical practice against the background of a relevant normative framework (as opposed to a not yet relevant normative background whose applicability should be investigated). Third, the theoretical background of this thesis (SDM) can be linked to the normative framework (SDM, autonomy, human dignity, and capacity of judgment), as both share the central premise of autonomous agents who are, in principle, capable of acting and have to be involved in decision-making in a developmentally appropriate way[37]. The normative framework is outlined in the following (figure 2).

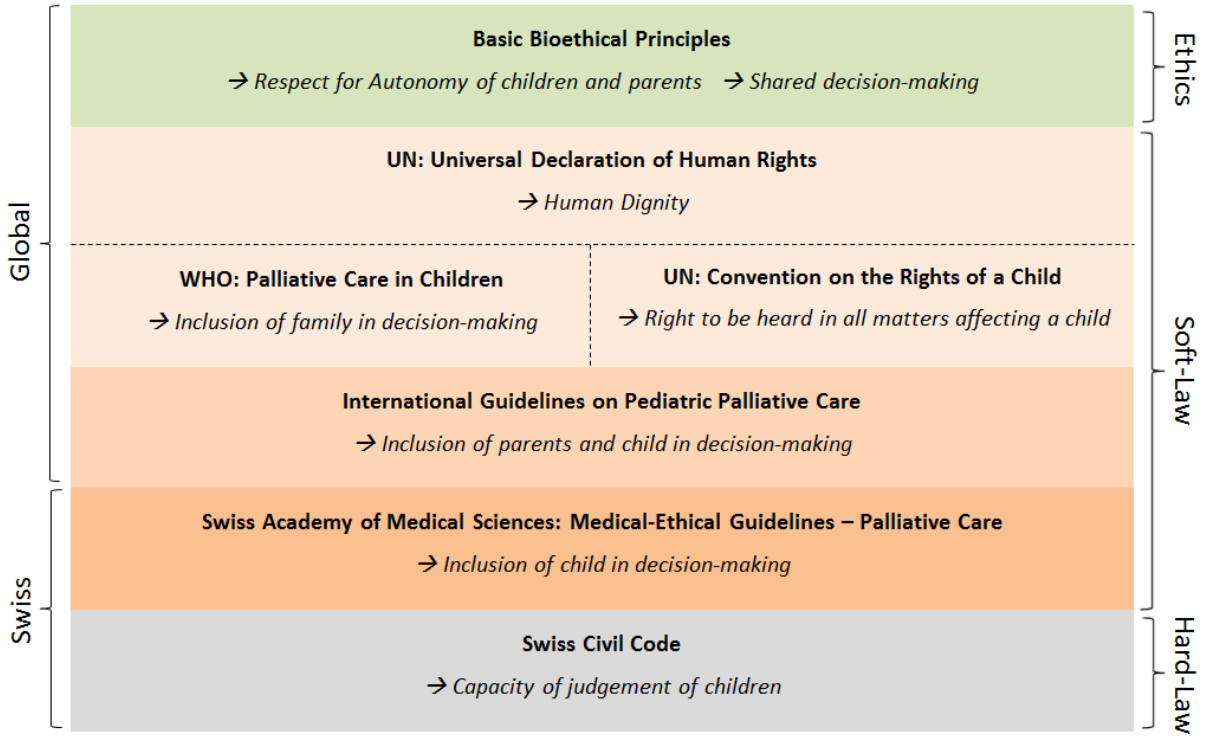
The respect for autonomy, one of Beauchamp and Childress' principles of biomedical ethics, has to be considered when analyzing SDM in pediatric oncology. Yet, as shown later, it needs to be complemented by relational aspects which it is failing to consider sufficiently[38]. According to the authors, the respect for autonomy demands the acknowledgment of "a person's right to hold views, to make choices, and to take actions based on personal values and beliefs"[39,p.63]. It is important to note that this respect should be translated into respectful action, not simply into a respectful attitude[39]. With regard to PPC, it is of crucial importance to enable children and their families to act autonomously (within a SDM approach), which entails facilitating reasoned and informed decisions. Moreover, two conditions need to be fulfilled in order to allow individuals to act in accordance with a self-determined intent: first, liberty (absence of controlling influences), and second, agency (being capable of intentional action)[39]. However, although Beauchamp and Childress's work is a landmark study on autonomy, they support a rather individualistic understanding of autonomy that disregards children's deep embeddedness into relationships. Such an individualistic understanding is closely linked to a cognitivist model of decision-making capacity. According to the cognitivist model, decision-

making capacity is as a mental trait residing within an individual mainly identified through logic and rationality[40,41]. Capacity assessment is based on the (mental) process that leads to the decision and is considered independent from the content of the decision[42]. The main critique regarding an individualistic understanding of autonomy is that it neglects the interdependence of the human self which, according to the critiques, always needs to be recognized as a situated and deeply embedded subject[38,43-46]. Also, the individualistic understanding of autonomy devalues persons who are highly dependent, such as children[38]. A more substantive approach to autonomy that takes non-cognitive determinants (e.g. social, cultural, emotional ones) into account is especially appropriate for children who are traditionally deemed incapable based on cognitive factors and who are emotionally, socially, and financially dependent on external factors[46,47]. Children's autonomy, their decision-making capacity, as well as their involvement are highly determined by their related-ness; children are embedded into relationships (with family members, professionals) on which they rely during the illness experience and also in face of decision-making, they are fundamentally inter-dependent. On the contrary, since the traditional individualistic understanding and the related cognition-based capacity assessment increase a child's likelihood of being excluded from decision-making, they hamper children's ability to participate in health-related decision-making and, thereby, reinforce their vulnerability. Therefore, doing justice to children requires broadening the perspective on autonomy by relational aspects and moving beyond merely applying an individualistic understanding.

Finally, from the recognition of respecting autonomy arises the imperative to share decision-making in a clinical setting, because SDM increases the chances for patient autonomy while guaranteeing both not to leave behind the patient as well as having the possibility to influence how the patient is benefited[27,48,49]. Moreover, SDM ultimately develops decisional capacity of a child who is guided and seconded in finding a good decision. Given this close relation

between autonomy and SDM, the respect for autonomy (in all its shades) marks a cornerstone when thinking about SDM in pediatric oncology.

Figure 2. Ethico-legal framework for shared decision-making



Note. UN = United Nations, WHO = World Health Organization.

Although not legally binding, the Universal Declaration of Human Rights is a highly influential document that represents a global consensus and has been further elaborated in numerous international, national, and regional legislations and policies[50]. The recognition of *every* human being as equal in dignity and as a subject of rights instantaneously leads to the derivation of fundamental human rights, such as the right to life, and with respect to PPC, to the derivation of autonomy and access to adequate medical care. Accordingly, a human being has the right to make decisions regarding his or her health, illness, and well-being.

Two other documents published by the United Nations offer a background against which SDM can be evaluated: the World Health Organization's "Cancer pain relief and palliative care in

children“ and the United Nations’ “Convention on the rights of a child”[5,51]. While the former recommends the inclusion of the family in decision-making, the latter grants the following rights to children. First, it grants the right to freedom of expression, which includes the “freedom to seek, receive and impart information and ideas of all kinds” and which shall be restricted only by law and if necessary (Art.13)[51]. Second, it grants the right “to express (…) [their own] views freely in all matters affecting the child”; furthermore, those views are weighted according to a child’s age and maturity (Art.12)[51]. Moreover, international guidelines on PPC unanimously recommend sharing decision-making with the child and the parents underscoring a developmentally appropriate involvement of children[1-3,7,29-34].

While the aforementioned documents claim transnational applicability, Swiss documents also must be taken into consideration when analyzing decision-making in the Swiss pediatric oncology setting. First, the Swiss Academy of Medical Sciences points out that the child-centeredness of PPC and involvement of the child have to be tailored to a child’s age, development, condition, and individual needs[52]. Further exemplifying the involvement of the child, the Academy underlines that minor patients can be capable of judgment with respect to treatment decisions, but capacity of judgement needs to be assessed for each situation and action present. This implies that a child’s capacity of judgement can vary across situations and actions and that fundamentally, children, under certain circumstances, may be the final arbiter in SDM. Finally, guidelines of the Swiss Academy of Medical Sciences are incorporated in the code of professional conduct of the Swiss Medical Association, which is binding for all its members, and a violation of the medical-ethical guidelines can be sanctioned by the Swiss Medical Association[53].

Second, Swiss legislation (Swiss Civil Code, Art.16) with regard to children’s decision-making rights does not distinguish between minor and major patients, but between patients with and

without capacity[4] of judgment. Minority per se does not deem a child incapable, it is a child's mental ability that determines whether or not the child is capable of judgement[54]. Thus, this decision-making capacity demarcates autonomy from dependency. It is generally assumed that adults are competent, but children as a class are usually considered incompetent with the underlying assumption that they also lack decisional capacity. Hence, decision-making capacity is a necessary criterion for legal competence: once a child is evaluated as capable of judgement he or she alone can legally consent to (or refuse) treatment[55]. For the medical context, minor patients between 12 and 18 years of age are mostly expected to be capable of judgement, but need to be evaluated on an individual basis[54]. Again, capacity of judgement therefore is relative with respect to time and the object in question. However, if a child is considered incapable of judgement (mostly assessed by physicians), his or her decision-making rights and autonomy are limited. In these cases, parents have to provide legally valid consent to treatments. Naturally, it is important to note that this Swiss legislation is legally enforceable and thereby different from principles of biomedical ethics, as well as from United Nations' documents and guidelines from medical associations.

From all the described documents, it is evident that normative premises can be derived for the purposes of decision-making in PPC. Thus, there is an espoused ideal of clinical practice that reflects how decision-making in pediatric oncology *ought to be*: building upon the respect for patients' autonomy decision-making should be shared. Presupposing a fundamental complementarity between the normative and the empirical, the question can be raised how this ideal of decision-making is translated into practice. Before that, however, it needs to be thoroughly outlined *how* the normative and empirical interrelate and how the chasm between them can be bridged. This meta-ethical reflection is undertaken in the following section.

[4] In this thesis, the term „capacity" refers to the cognitive decision-making ability within the clinical setting. The term "competence" describes a person's authority to translate such decisions into legally binding ones.

2 Empirical Bioethics Methodology – Bridging the Empirical-Normative Divide

As outlined in the previous section, decision-making in PPC is neither an ethical nor a legal adiaphoron but rather a highly normatively loaded concept. Since this thesis aims to empirically study decision-making in PPC and to integrate the empirical data with normative ethical analysis, it is of preeminent importance to a priori think about how empirical research can be integrated with normative bioethical reflection.

2.1 Empirical Bioethics

Shifting away from a purely theoretical discipline and thereby gradually relinquishing its autonomy regarding the evaluation of ethical issues in the medical sphere to other disciplines, bioethics has been incorporating social scientific empirical methods for more than three decades[56]. This so called "empirical turn" usually describes the steadily growing number of empirical research studies to inform normative approaches in bioethics[57]. Almost twenty years ago, Birnbacher argued that empirical research does not supplement (applied) ethics, but complements it and ultimately calls for a co-operation of social sciences and ethics[58]. Critizising traditional ethical approaches that risk neglecting the realities of lived experiences, empirical bioethics generally underlines the necessity of utilizing social sciences in order to become more contextually aware[59-62]. Although different reasons for using empirical data exist among ethicists, all empirical bioethics approaches – as a starting point – deny the incompatibility of empirical data and moral theory and conceive both as complements[58,63,64]. By way of exemplification, factual statements about how the world *is* (derived from quantitative or qualitative methods) are seen as compatible with normative statements about

how the world *ought to be* (based on practical ethical reasoning), or to put it more concisely: empirical and normative analyses converge in the field of empirical bioethics.

According to Leget, "empirical bioethicists use qualitative, quantitative or mixed methods to gather data relevant to judgements concerning moral matters in medicine and medical science."[65,p.228] Moreover, there are basic assumptions underlying any sort of empirical ethics: (a) studying an actual practice provides meaningful information for the bioethical endeavor which, in any case, should begin from this particular premise, (b) social sciences' methodology is well suited to map this reality, (c) empirical and normative approaches are compatible and fundamentally complementary (d) empirical ethics, in its overall meaning, is not a methodology of doing ethics, but a methodological attitude regarding the use of empirical research in ethical reflection, (e) empirical ethics is not an anti-theorist approach, in which the empirical is the final moral arbiter[66].

Why conduct empirical bioethics?

If there were no benefits of conducting empirical research on PPC in order to improve practice, solely working from a normative standpoint would suffice. Nonetheless, empirical bioethics is not an end in itself, but needs to be justified. De Vries and van Leeuwen give the following five practical reasons for using empirical data[67]. First, by including moral experiences and relevant expertise of practitioners in ethical reasoning, relevant aspects of the medical practice (e.g. internal norms, practical wisdom, context-driven paths for putting abstract principles into practice) can be better understood. Second, the particularities of medical practice (e.g. inpatient-settings, minors' possible incapacity of judgement, triadic decision-making constellation, physicians' attitudes) should be considered in order to familiarize bioethics with daily life within a certain setting. Third, the likelihood of successfully implementing the results of ethical reasoning is increased if special characteristics of the medical practice are taken into consideration. Fourth, the ethical analysis is strengthened by acknowledging moral dilemmas

that occur in medical practice. Fifth, empirical data decreases the odds for biased conclusions and self-justification on the part of the researcher or the research team. Further, without empirical input, it remains unclear on which norms the normative judgment should be based. The normative becomes disconnected from "lived morality", which, however, has to be considered because many normative questions cannot be answered due to a lack of a universally shared normative framework[65]. As follows from the previous considerations, empirical bioethics represents an indispensable step towards drawing normative conclusions through the integration of empirical and normative analysis[61]. Without referring back to empirical analyses, bioethics to a large extent forfeits its ecological validity, which means the degree to which its concepts and norms are applicable in practice.

How to conduct empirical bioethics?

With regard to practical research strategies for empirical bioethics, numerous typologies of how to bring together the empirical and the normative have been put forward[59,60,63,65,68]. Some authors have distinguished different relations between the empirical and the ethical spheres[59,68]. Kon presents four hierarchically ordered levels on which empirical research informs bioethics[68]. On the bottom level – the lay of the land research – studies describe and reveal the current practices, opinions, beliefs or other aspects of the status quo. On the second level – ideal versus reality – studies explore to which extent actual clinical practice matches with an espoused ideal. Upon this builds the third level – improving care – where studies examine how practice can be made more consistent with ethical norms. Finally, on the fourth level – changing ethical norms – based on the work of the previous three levels, amendments of ethical norms are recommended. Although to different extents, this thesis' empirical work informs bioethical considerations on SDM within the context of PPC on all four levels.

Three categories (containing ten subcategories) of how the *is* can contribute to the *ought* are sketched by Solomon: (a) facilitating the move from ethical analysis to ethically justifiable behavior, (b) enhancing ethical analysis and justification, and (c) generating new normative concerns[59]. The first and the last category offer subcategories that capture the role of empirical analysis within this thesis: documenting gaps between ideal and actual practice, and describing the environmental context that mediates moral action (subcategories of the first category), as well as identifying new moral problems, specifying acknowledged problems, and clarifying causal mechanisms (subcategories of the third category)[59].

In a different and more practical strategy that depends on what – theory or data or both – is the final moral arbiter (e.g. locus of moral authority), Molewijk et al. distinguish five different research strategies of how to arrive at a normative conclusion (figure 3)[63]. First, in *prescriptive applied ethics* moral theory is the final moral arbiter and empirical data cannot cause substantial adjustments of a moral theory, if it does not concur with the morality of a particular practice (one way interaction: top down between theory and data). Second, *theorists* use empirical data in order to improve or refine a moral theory (one way interaction: bottom-up between data and theory). Third, according to *particularists* the morality within a particular practice is the only moral arbiter (e.g. casuistry; no interaction between theory and data). These three strategies lack a two-way interaction that actually brings theory and data together. The following two strategies offer insights into how to draw normative conclusions by integrating moral theory and empirical data. The fourth strategy – *critical applied ethics* (CAE) – does not accept moral authority of either theory or data, instead it simultaneously lets data criticize theory and applies critical thinking of ethics to the data throughout a fivefold process (two-way interaction). Finally, *integrated empirical ethics* overcomes the theoretical hybridization of normative and empirical disciplines, stresses instead that the empirical and the normative are mutually constitutive, and integrates theory and data through intensive cooperation between ethicists and

descriptive scientists in order to reach normative conclusions (no interaction, since normative and empirical fall together).

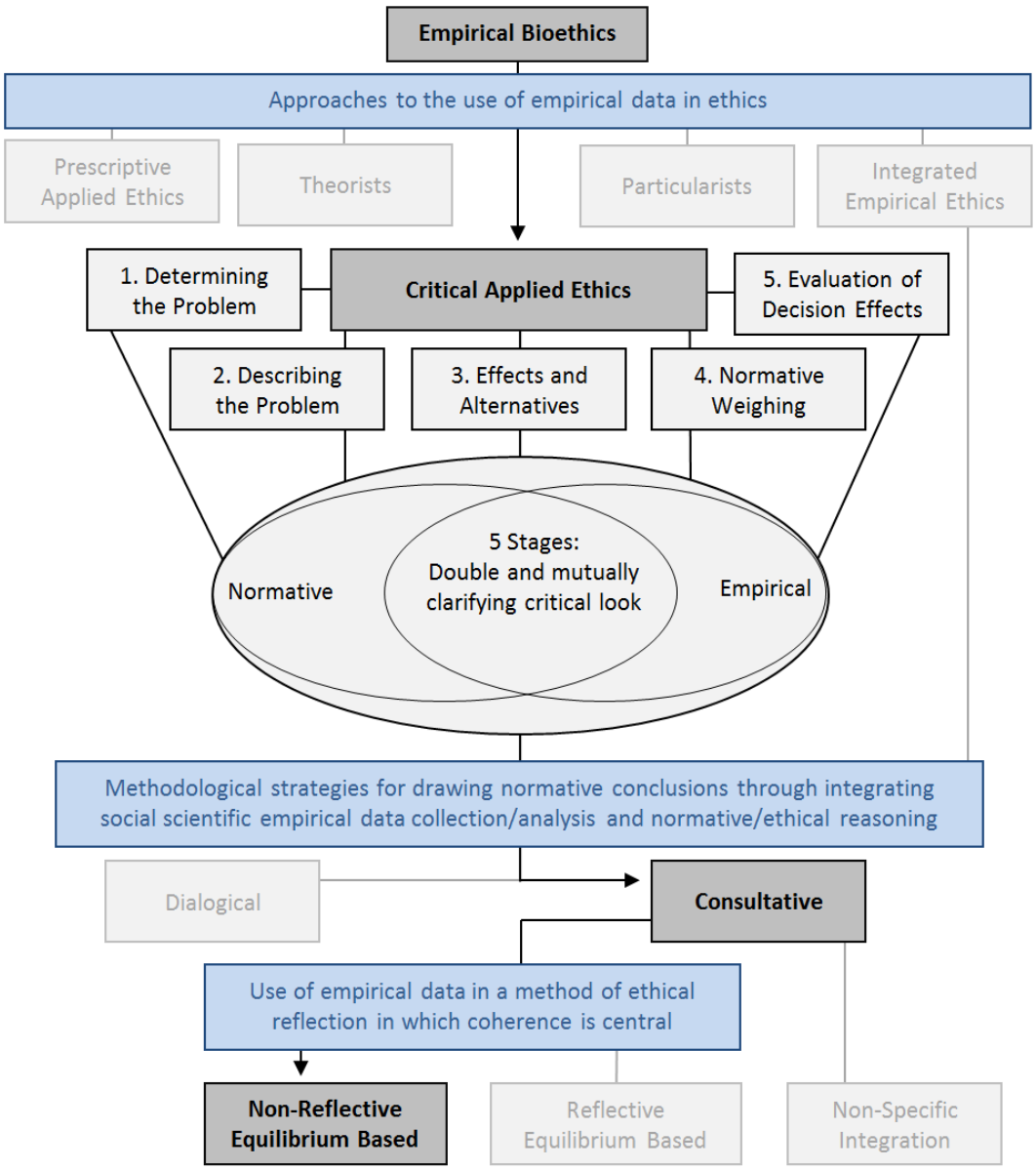
The outlined typologies of relating the empirical and ethical spheres are valuable as they shape the field of empirical bioethics in terms of the aims of (Kon[68], Solomon[59]) and in terms of the moral authority (Molewijk et al.[63]) within a particular research project. In short, they represent ways of thinking about the research project, however, they do not (fully) dig into details and differences of the different methodologies within each typology[60]. Therefore, the approach to integrating ethical theory and empirical data as applied in this thesis is carved out in detail in the following.

2.2 Approach to Conducting Empirical Bioethics in this Thesis

Out of the introduced five methodologies by Molewijk[63], the first three do not intend any two-way interaction between normative and empirical and, because of that, are not sharing this thesis' view of a mutual co-determination of the normative *and* the empirical. Whilst integrated empirical ethics (fifth strategy) might be an appealing approach, Leget and colleagues' criticism contains a valid point that, taken seriously, renders the latter less appropriate for bioethical research as compared to CAE (fourth strategy)[65]. They argue that integrated empirical ethics is "both conceptually contradictory and methodologically impaired": conceptually because it cannot but draw on the distinction between fact and value when explaining how the postulated empirical-ethical-hybrid should be formed, thereby referring back to the distinction they seek to overcome, and methodologically, since the critical interdependence between social science (that could not be critically explored with regard to its normative dimensions anymore) and normative ethics (that could not be studied by empirical researchers anymore) dispersed[65,p.231]. Consequently, in order to draw normative conclusions through integrating the empirical and the normative this thesis' approach draws on CAE as

established by Leget et al.[65,69], but reaches beyond this methodology by enriching it through further methodological in-depth considerations as suggested by Davies et al.[60] (figure 3).

Figure 3. Empirical bioethics approach



Note. This figure was synthesized based on the works of Molewijk et al., Leget et al., and Davies et al.[60,63,65]

CAE uses empirical data for a continuous, fivefold process (figure 3) of reassessing and refining the normative[60,65,69]. At all five stages, CAE appreciates the normative and the

empirical as two co-determining poles of the bioethical ellipse which offer a double and mutually clarifying critical look to the issue at stake (figure 3)[65].

CAE begins with *the determination of the problem*, either in normative ethics or in empirical research. Once a moral problem is identified at the first stage, it has to be *accurately described* at the subsequent stage, again, from both perspectives: the normative ethics analyses the (sometimes hidden) values that are inherent to the categories used in the determination of the problem, while empirical research carefully studies how the respective practice actually looks like. At the third stage, *effects and possible alternatives* to envisaged actions become relevant. Not only effects (e.g. does training for practitioners lead to better care outcomes?) can be assessed by empirical research, also premises of normative arguments can be tested (e.g. whether children actually want to be involved in decision-making). However, the function of empirical data at this stage is limited, in particular when the moral issues are essentially non-consequentialist and normative ethics refers to deontological approaches as well as to the possibility of a morality that is supported by ethical theory, but not by data. At the fourth stage – *the normative weighing* – the normative and empirical are densely amalgamated in critical reciprocity. Most importantly, ethics monitors that the normative power of the descriptive is evaluated properly. Empirical research, in turn, has to critically question the ethical theories that are utilized. Since bioethics needs a constant evaluation of its outcomes, the *effects of a decision need to be evaluated* at the final stage. Empirical research explores the effects, while ethicists reflect upon the effects and ensure that consequentialism is not – unreflected – triumphant over possibly more appropriate theories.

In their systematic review of empirical bioethics methodologies, Davies and colleagues seek to highlight in detail how normative conclusions are drawn based on the integration of social scientific empirical data collection/analysis and normative/ethical theorizing[60]. They identify two poles of a continuum of methodological orientation, namely *dialogical* and *consultative*

methodologies (figure 3). While the former class revolves around a close dialogue between stakeholders and researchers to reach, first, a common understanding, and, second, a commonly drawn conclusion in face of a discrete problem, CAE falls within the latter class of consultative methodologies. Consultative approaches rely on an external "thinker" (e.g. research group, individual researcher) who analyses data and develops normative conclusions after and independently of the data collection. In this way, participants are consulted to obtain their attitudes and experiences, but are not involved in data analysis and ethical reflection.

Consultative methodologies can be further sub-categorized based on the process by which the "thinker" forms a normative conclusion[60]. Three forms of non-specific integration (bottom up, top down, equal weighing) and two coherence seeking methodologies can be distinguished: reflective equilibrium based (employing one of numerous versions of the precisely defined methodology *reflective equilibrium*[60]*)* and non-reflective equilibrium based (not employing the particular methodology *reflective equilibrium*, although still reflective in nature) with CAE being part of the latter. In this regard, CAE seeks coherence, thereby localizes moral authority in rationality and consistency, and balances the relevant and equally weighted considerations until a coherent position is found[60].

Finally, Davies et al. recommend to thoroughly think through three central questions within any empirical bioethics endeavor, namely regarding the justification of a normative conclusion (through consensus or coherence), the analytic process that leads to the conclusion (prioritizing the thinker, theory, or data), and the kind of conclusion (aiming for particularity or generalizability)[60]. In short, this thesis seeks for coherence, prioritizes the "thinker" (in the form of an interdisciplinary research team), and aims to evaluate and enhance a particular medical practice, namely decision-making regarding PPC in pediatric oncology.

3 Research Objectives and Empirical Methods

3.1 Research Objectives

This thesis is part of a larger mixed-method research project titled "*Attitudes and motives concerning end-of-life decisions: Competency and autonomy of children and adolescents in paediatric oncology*". The first prospective part of the project targeted children in their decision-making processes throughout the whole course of illness and treatment. The prospective study gathered data from children (interviews), parents (interviews, questionnaires), and physicians (interviews, questionnaires), but could not gather sufficient information on decision-making regarding palliative care; because of two main reasons. First, the sensitive nature of the topic might have withheld families of participating in the study. Second, (fortunately) only few children (7 out of 21) were considered palliative. Hence a follow-up study was designed to retrospectively understand the end-of-life situation and to examine how and by whom palliative care decisions are made.

Unlike curative treatment decision-making regarding palliative care is not guided by protocols and thus lacks a universal approach. Further research on the involvement of children as well as on the dynamics of the triadic constellation of decision-making is needed[26,70]. Thus, the goal of this thesis is to propose ethically sound and practically feasible ways to facilitate SDM regarding palliative care in the context of pediatric oncology. To fulfil this objective, this thesis aims to answer the following research questions.

1. *What are parents' and oncologists' perceptions of children with respect to their involvement in decision-making? (chapter 2)*
2. *Which factors determine parents` and oncologists` decisions to respect or limit a child's role in decision-making? (chapter 2, 3)*
3. *Which additional factors influence shared decision-making? (chapter 3, 4, 5, 6, 7, 8, 9)*

4. *What are stakeholders' attitudes towards and experiences with the involvement of children in decision-making? (chapter 5)*
5. *How are decisions regarding the transition from curative to palliative care structured and made? (chapter 3, 6)*
6. *What are barriers to the implementation of pediatric palliative care and what are possible strategies to overcome them? (chapter 5, 7, 8, 9)*
7. *How do international guidelines and practitioners understand PPC? (chapter 6, 7, 8)*

Referring back to Kon's four levels on which empirical research can inform bioethics, it can be noted that the presented research questions apply to all four levels[68]. Accordingly, this thesis seeks to define current medical practice, to assess the congruency of the "is" and the "ought", to improve care and to recommend changes in the respective normative background.

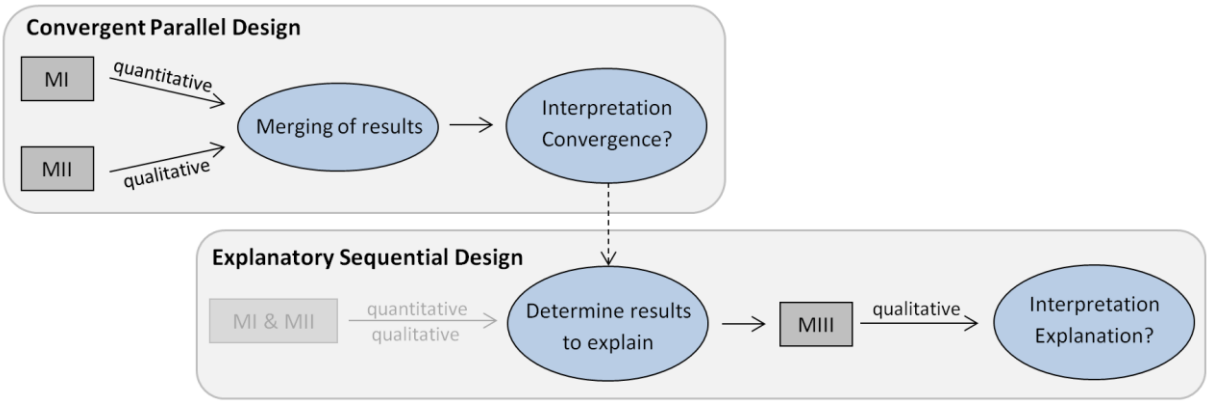
This thesis has its origin in the realization that SDM in pediatric oncology needs further improvement and is still understudied. According to a recent systematic literature review, further research that identifies factors contributing to favorable participation of the child in decision-making regarding palliative care is needed[70]. Furthermore, despite recommendations by international guidelines[2,7,33] and the findings of the first phase of the overall project which indicate that children value to be included in decision-making[71,72], children's participation is often still limited [70,73-77]. Also the actual parental roles in decision-making were shown not to match the preferred roles with the former being too passive[78]. Moreover, with regard to PPC, studies show that there are still high numbers of late or non-referrals which do not meet recommendations to start PPC at diagnosis[74,79-82]. Finally, practitioners lack formalized training in PPC and often need to rely on trial and error learning[74,83,84].

3.2 Empirical Methods

This thesis followed a mixed-methods design. The particular methods are described in detail in the methods sections of the articles below and, therefore, are not presented here. However,

it is crucial for the reader's comprehension to outline how these different qualitative and quantitative methods were combined by briefly presenting this thesis' mixed-methods approach. The empirical research in this thesis was comprised by two mixed methods designs: a convergent parallel design and an explanatory sequential design (figure 4)[85,86].

Figure 4. Mixed methods design



Note. M = Module.

Quantitative results of surveys completed by physicians and parents of children diagnosed with cancer (stemming from the first prospective part of the project) as well as quantitative results of extractions of medical records of deceased children represented the first quantitative module. Concurrently, guidelines on PPC were analysed. This represented the first qualitative module. Subsequently, the results of module I (quantitative) and module II (qualitative) were merged, analysed, and interpreted. In this phase, the researchers were able to determine the extent to which the results of the two modules converged. Furthermore, these results and interpretations informed module III (qualitative), namely focus group interviews with different stakeholders of decision-making regarding PPC. In this phase, the researchers addressed follow-up questions to experts and, thereby, reached a deeper understanding of the issue through an explanation of the previous results.

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Chapter 2

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Parents' and Physicians' Perceptions of Children's Participation in Decision-making in Paediatric Oncology: A Quantitative Study

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Abstract

Objective: The goal is to present how shared decision-making in paediatric oncology occurs from the viewpoints of parents and physicians.

Methods: Eight Swiss Pediatric Oncology Group centres participated in this prospective study. The sample comprised a parent and physician of the minor patient (<18 years). Surveys were statistically analysed by comparing physicians' and parents' perspectives and by evaluating factors associated with children's actual involvement.

Results: Perspectives of ninety-one parents and twenty physicians were obtained for 151 children. Results indicate that for six aspects of information provision examined, parents' and physicians' perceptions differed. Moreover, parents felt that the children were more competent to understand diagnosis and prognosis, assessed the disease of the children as worse, and reported higher satisfaction with decision-making on the part of the children. A patient's age and gender predicted involvement. Older children and girls were more likely to be involved. In the decision-making process, parents held a less active role than they actually wanted.

Conclusion: Physicians should take measures to ensure that provided information is understood correctly. Furthermore, they should work towards creating awareness for systematic differences between parents and physicians with respect to the perception of the child, the disease, and shared decision-making.

Introduction

Decision-Making in Paediatric Oncology

When children are diagnosed with cancer, families and physicians face the cumbersome task of making urgent and difficult treatment decisions. In the paediatric setting, decision-making process includes multiple steps and at least three parties: the physician/nurse, the patient, and the parents: each with their own opinions, needs, and expectations[1]. They form a triadic constellation that must share the process and make a decision in the best interest of the child. Literature on shared decision-making emphasizes the following aspects: a) the involvement of at least two parties; b) sharing of information between the parties; c) consensus regarding the preferred treatment; and d) successfully achieving an agreement[2,3]. Shared decision-making requires the involvement of all parties, with the child participating in a developmentally appropriate way[4]. However, neither the participation of the child nor the ability of parties to carry out their preferred role is guaranteed. For instance, Mack concluded that more than one-third of the parents held a passive role and that they were unsatisfied with the information they received[5,6]. Moreover, physicians often face several obstacles to communication such as time limitations and uncertainty about the patient's current or projected condition[7,8]. Finally, despite recommendations by international guidelines to involve children[9,10] several studies have noted that children's participation is still low and that they are often shielded from difficult or bad information[11-14].

Factors Hindering the Decision-Making Process

Forming a shared decision is not an easy process since all parties must overcome several difficulties. First, factors that inhibit parents include coping with the possible loss of their child and its consequences for the family[15]. Parents must overcome intra-familial conflicts,

may have unrealistic expectations regarding cure, and may deny that the cancer is terminal[16]. Parents' limited understanding of the medical information and low family educational level also impair their ability to adequately take part in the decision-making process[17].

Second, physicians perceive a series of ethical challenges in making treatment decisions. These include weighing what the consequences of their actions would be, questioning the role of parents, and uncertainty as to how the child's wishes should be considered[7]. Physician's wishes to maintain some degree of hope may result in avoiding frank disclosure, thereby hindering decision-making. Furthermore, they face difficulties when asked "to provide uniquely tailored, culturally appropriate, holistic, comprehensive, coordinated, long-term care to all families"[6,18,19]. These concerns become more burdensome in light of the little formalized training that physicians receive in paediatric palliative care and in light of their reliance on learning through trial and error[13,16,18].

Study Purpose

Available literature illustrates the need to shift the actual decision-making towards a process that empowers every involved person to occupy their preferred role. To know more about how shared decision-making in these situations occurs and how children are involved, more studies are needed. This research gap was addressed in this study carried out with physicians working in Swiss Pediatric Oncology Group (SPOG) centres and parents of children suffering from cancer. Study participants were questioned about their attitudes towards the child's participation in the decision-making processes, their satisfaction with the process, and the actual involvement of the child. The study posed the following research questions: What are parents' and physicians' attitudes and orientation regarding inclusion of children in their cancer treatment decisions? What are their opinions on several aspects of shared decision-making and do they differ? Which factors determine children's actual involvement?

Methods

Study Design

Eight of the nine SPOG centres in Switzerland participated in this multicentre mixed methods project. The qualitative part of the project included interviews with children, their parents, and physicians. The results from the qualitative interviews have been reported elsewhere[20-23]. In addition, a quantitative collection of information using closed-ended surveys took place at the participating SPOG centres. In this quantitative part, children were not included. In this paper, we report the results of the quantitative surveys completed by parents and physicians. Distribution of the surveys began in November 2012 and was carried out until April 2015. Ethical approval was obtained from the responsible ethics committees for each SPOG centre. This inevitably meant that data could not be collected at all centres at the same time. The surveys were completed on a rolling basis according to when we received the ethics approval. The first centre began distributing the surveys and collecting them in November 2012 and the last one in June 2013. All centres ceased data collection in April 2015.

Study Population

Parents and treating physicians were included in the quantitative part of the project, if the respective child (a) was less than 18 years of age and (b) had a cancer diagnosis and received cancer treatment in one of the participating SPOG centres. The views of the paediatric patients were not gathered because we could not be sure that young children (less than twelve years) could understand and complete the study survey correctly. However, some variables captured children's views indirectly through the parents or the physicians evaluation of the child's view (e.g. "How satisfied was your child with decision-making?"; "Please evaluate your child's suffering due to the disease").

Data Collection

Before starting data collection, the research team visited the respective SPOG centres to introduce the study, its methodology, and study tool to the physicians, as well as to the data manager (where possible). The purpose of this visit was to explain the recruitment process so that data collection would be as uniform as possible within each centre and between different centres. Study materials with codes for physician and parent were labelled for each patient by the researchers and delivered to the participating centres. The data manager or the responsible contact person for the centre kept a note on which participant received which code. To ensure confidentiality, the researchers did not have access to participants' identifiable information.

The study team requested each physician at the participating centre to complete one survey for every patient he or she treated. This meant that the physicians completed multiple surveys; however, each was for a unique patient case. They were also asked to approach the parents for each patient for whom they filled out a survey. The treating physician thus informed the parents about the study and provided the parents the study information documents: informed consent, a survey, and refusal card. Based on their preference, the parents could either return the survey to the hospital in a sealed envelope or post it using the self-addressed stamped envelope provided. Since parents completed the survey within a short time span of a few weeks after they were approached by their child's treating physician, we expect that within one dyad perspective, the point along the child's disease trajectory (e.g. diagnosis, relapse) would not have differed greatly. By emphasizing that parents have the opportunity to refuse to participate and by handing over a refusal card, the study team ensured that no undue pressure was placed on parents, given their difficult situation.

Study Sample

A total of 229 surveys were completed and returned (138 by twenty treating physicians; ninety-one by parents) during the data collection period. These 229 responses represented 151 unique children cases. From the 151 children, dyad-perspective (of parent and physician) was captured for seventy-eight children. For seventy-three children, only one perspective was available: sixty from the treating physician and thirteen children from a parent. We cannot confidently estimate the number of patients who sought treatment at the participating SPOG centres during the study period as this data is not obtainable for the research team. However, twenty of the twenty-eight physicians at the participating SPOG centres participated in the study. Since 138 surveys were completed by the twenty physicians, we expect that 138 parents received a survey. From those parents who have received a survey, a completed survey was sent to the research team in 66 per cent of the cases. We received a total of eleven refusals from the parents.

Study Questionnaire

The study tool focused on the inclusion of children in the overall treatment decision-making. Several aspects and items of the detailed questionnaire were developed from the research team's knowledge in the field and input from collaborating physicians. The survey was designed to gather the following data: a) demographics information; b) the amount of information given to the parents and whether the patient was present at this time; c) the capacity of the patient to understand disease-related information; d) decision-making and satisfaction with decision-making within the triadic system of child, parent, and physician; and e) current and preferred role of parents within decision-making. Questions concerning roles in decision-making were adapted and revised from Mack and colleagues. The questionnaire consisted of items with categorical responses or Likert scales. It was pilot

tested in August 2012 in one SPOG centre. A few adaptations were made that did not change the questionnaire's overall purpose.

Statistical Analyses

A research assistant entered all completed surveys into SPSS 22 and another checked for correctness of data entry. Statistical analyses were performed using SPSS 22 (SPSS Inc, Chicago, IL). For analyses described below, reported *p* values are two-sided and statistical significance level was set at $p < .05$.

To understand the general age at which children are considered capable of understanding different treatments and related consequences, physicians' evaluations of the age from which the majority of children were considered able to understand various information related to their illness and capable of making related decisions were assessed descriptively. To be able to determine this age, we first counted how many children at a given age were considered capable versus how many children of the same age were not. Second, we examined the age at which these frequencies shifted from "more children were deemed not capable" to "more children were deemed capable." This shift represented the "turning point" that we describe in this paper.

Moreover, we compared physicians' and parents' perspectives on the decision-making process, on children's characteristics, and on disease-related features. Using the seventy-eight dyad-perspectives a Wilcoxon signed-rank test was carried out to evaluate differences between physicians' and parents' responses to the following seven variables: suffering of the child, prognosis of child's cancer, capacity of the patient to understand disease-related information, past and expected treatment duration, satisfaction with decision-making, current and preferred role of parents in decision-making, and amount of information given to the

parents. Additionally, using the parental perspective, we compared parents' current and preferred role in decision-making in order to evaluate whether they hold the role they wanted. Finally, we evaluated factors associated with the actual involvement of the child in the shared decision-making process using generalized linear mixed model (GLMM). Categorical responses regarding the involved parties in decision-making (question: "who was involved in decision-making?") were dichotomized into "with child" and "without child." This binary variable was the dependent variable. Based on a priori theoretical considerations, four predictor variables were included: age of the child, gender of the child, cancer prognosis, and physician's professional experience as a paediatric oncologist. Since children receiving care from a particular physician and/or centre might have similar data, the analysis was adjusted for clustering within physicians and SPOG centres. The GLMM analysis included the 138 cases that were completed by twenty physicians.

Results

Demographic Characteristics of the Sample

Of the children, 62 per cent were male. Parents were between eighteen and fifty-nine years old, and most of them were mothers (80 per cent; two missing values). Physicians were between thirty-five and fifty-eight years old, with a small majority (56 per cent) being female (two missing values). Other demographic information of patients, parents, and physicians is presented in table 1.

Table 1. Descriptive statistics of children and study population

Children (n=151)[1]		Parents (n=91)		Physician (n=20)	
Age (M; *SD*)	8.05 (4.85)	Age (M; *SD*)	39.16 (7.23)	Age (M; *SD*)	43.56 (6.28)
Gender (male)	62%	Gender (male)	16%	Gender (male)	44%
Prognosis (M; *SD*)	1.63 (0.93)	Nr. children (M; *SD*)	2.22 (0.93)	Experience (<8 years)	50%
Suffering (M; *SD*)	2.49 (1.00)	Religious (yes)	54%	Religious (yes)	39%
Prev. Treatment (<6 months)	63%	Marital Status (married)	82%		
Exp. Treatment (<12 months)	31%	Relationship with the child (father)	16%		

Note. Prev. = Previous; Exp. = Expected.

[1] Information about children was obtained from physicians and parents. Due to minor discrepancies between parents and physician, we evaluated age and gender for the ninety-one cases from parents and the remainder from physicians. Prognosis, suffering, previous treatment, and expected treatment were evaluated by the physicians in all cases, except for thirteen cases for whom we did not have the physician survey. Prognosis was measured by a five-point Likert item ranging from 0 ("extremely good") to 4 ("very bad"); suffering from 0 ("a great deal") to 4 ("not at all").

According to the twelve categories (I–XII) of the International Classification of Childhood Cancer (ICCC), the most frequent diagnoses were as follows: leukaemia (ICCC-I; 49.7 per cent), central nervous system neoplasms (ICCC-III; 18.5 per cent), malignant bone tumours (ICCC-VIII, 7.9 per cent), and lymphomas and reticuloendothelial neoplasms (ICCC-II, 6.6 per cent). Two diagnoses were not represented in our sample: retinoblastoma (ICCC-V) and hepatic tumours (ICCC-VII). Compared to the Swiss Childhood Cancer Registry (SCCR), leukaemia was over-represented (49.7 per cent vs 33 per cent) and central nervous system

neoplasms were comparable (18.5 per cent vs 19.6 per cent) (Swiss Childhood Cancer Registry 2016). Patients' ages were overall comparable to SCCR (in brackets): 0–4 years 34.8 per cent (36 per cent), 5–9 years 26.2 per cent (21.5 per cent), 10–14 years 27.5 per cent (22.7 per cent), and 15–20 years 11.4 per cent (19.8 per cent; note: SCCR includes adolescents up to twenty years of age).

Physicians' Evaluations of Children's Understanding and Capacity

With regards to understanding diagnosis, only one out of four children who were five years of age were deemed capable, three out of eight children who were six years of age were considered capable, and the same goes for seven out of thirteen children who were seven years old, and seven out of nine for children eight years old. Accordingly, the turning point was reached between six and seven years of age (table 2).

*Table 2***.** Turning points[1] of children's competency evaluations by physicians (n=138)

Variable	Age (years)
Understanding diagnosis	6.5
Understanding prognosis	9.0
Understanding cancer cause	9.5
Understanding response to treatment	6.0
Making treatment decisions	11.5
Making decisions to be included in CT	11.5

Note. CT = clinical trial.
[1] From this age physicians considered the majority of children at a given age capable of understanding/ decision-making.

Therefore, physicians judged understanding of response to treatment and understanding diagnosis to be easiest and thus deemed the majority of children older than six years to be capable of these two tasks. Understanding of cancer cause and prognosis was reported more positively for those children who were nine years and older. The capacity to make treatment-related decisions was evaluated as most challenging with the age limit for these choices being

above eleven and a half years. Because of lower numbers we do not present the evaluations of the parents.

Factors Influencing Decision-Making Process

With regard to the *provision of information* the results highlight that for all six aspects of information provision (diagnosis, prognosis, treatment options, cancer cause, response to treatment, and clinical trial inclusion) parents' and physicians' perceptions differed significantly (table 3). Compared to physicians, parents rated the amount of information that was given to them by the physicians as being less satisfactory.

Second, concerning *children's understanding of disease-related information*, results indicate that parents evaluated children's ability to understand diagnosis and prognosis higher than how it was evaluated by the physicians. Parents thus had a more capable image of their children (table 3). Regarding the *characteristics of disease*, parents' and physicians' ratings of the suffering of a child as well as the expected treatment duration differed significantly. Parents assessed the disease of their child as worse (higher suffering, longer duration) than how physicians evaluated the disease. Finally, concerning *satisfaction with involvement in the decision-making process*, parents rated a child's satisfaction with the actual decision-making as higher than the physician (table 3).

Parents' Preferred and Current Role in Decision-Making

Study results present that parents held a less active role than they actually wanted, $Z = -3.080$, $p = 0.002$. Of the parents who reported both their current and preferred role, 64 per cent reported that their current roles matched their preferred role; 8 per cent reported a more active role, and 28 per cent a less active role. In order to further examine this difference in current and preferred roles, an exploratory GLMM analysis was performed addressing the

question of what determines parents' less active role. This analysis did not reveal any predictors.

Table 3. Wilcoxon signed-rank test comparing physicians' and parents' perceptions on elements related to decision-making (dyad-perspective n=78)

Variable	PH=PA	PH>PA	PA>PH	z	p
1. Provision of information					
Information Diagnosis[a] (n=62)	48	13	1	-3.116[i]	.008[j]
Information Prognosis[a] (n=63)	39	20	4	.3.563[i]	.000[j]
Information Treat. Options[a] (n=62)	43	16	3	-3.065[i]	.006[j]
Information Cancer Cause[a] (n=62)	27	22	13	-2.178[i]	.029[j]
Information Response to Treat.[a] (n=62)	44	16	2	-2.840[i]	.010[j]
Information Inclusion CT[a] (n=56)	41	14	1	-3.231[i]	.005[j]
2. Children's understanding of disease related information					
Understanding Diagnosis[b] (n=67)	27	13	27	-2.202[h]	.028[j]
Understanding Prognosis[b] (n=63)	29	11	23	-2.497[h]	.026[j]
3. Children's competency to make treatment related decisions					
Competency Treatment Decisions[b] (n=66)	33	15	18	-.553[h]	.580[j]
Competency CT Decisions[b] (n=64)	31	15	18	-1.295[h]	.390[j]
4. Characteristics of disease					
Prognosis[c] (n=61)	20	21	20	-.162[h]	.872[j]
Suffering of the Child[d] (n=62)	27	10	25	-2.830[h]	.010[j]
Expected Treat. Duration[e] (n=67)	45	5	17	-2.802[i]	.015[j]
5. Satisfaction of involved parties					
Satisfaction Child[f] (n=33)	12	6	15	-2.538[h]	.011
Satisfaction Parent[f] (n=62)	23	15	24	-1.711[h]	.087
Satisfaction Physician[f] (59)	34	9	16	-1.224[h]	.221
6. Shared decision-making					
Shared Decision-making[b] (n=65)	16	20	29	-1.239[i]	.215
Preferred Role of Parents[g] (n=65)	29	22	14	-1.171[h]	.242

Note. CT = clinical trial; Treat. = treatment; PH=PA = physicians and parents rated equally; PH>PA = physicians rated higher than parents; PA>PH = parents rated higher than physicians.
[a]five-point Likert item ranging from "full information" to "no information"; [b]five-point Likert item ranging from "absolutely agree" to "strongly disagree", [c]five-point Likert-item ranging from "excellent" to "very bad"; [d]five-point Likert-item ranging from "severe" to "no suffering"; [e]four-point Likert-item: "less than one year", "between one and two years", "between two and four years", "more than four years"; [f]five-point Likert-item ranging from "very satisfied" to "not satisfied"; [g]seven-point Likert-item ranging from "I prefer to make the decision with no input from the physician" to "I prefer that the physician makes the decision with no input from me"; [h]based on negative ranks; [i]based on positive ranks; [j]since the overall-hypothesis was tested through multiple comparisons and to control for the increased likelihood of a type I error, p-values were adjusted applying Bonferroni-Holm correction.

Characteristics of Children Involved in Decision-Making

Only forty-four (out of 137) children were involved in decision-making. They belonged to these age groups: three out of fifty children from zero to four years, six out of thirty-six

children from five to nine years, twenty-three out of thirty-eight children from ten to fourteen years, and twelve out of thirteen from fifteen to seventeen years. The findings from the GLMM reveal that a patient's age and gender significantly predicted whether the child was involved or not (table 4). In particular, the older a child, the more likely was his or her involvement. Also, girls were more likely to be involved than boys. To illustrate, an additional year in age resulted in higher odds of being involved by a factor of 1.7; for a girl instead of a boy, the odds increase by a factor of 3.7. An exploratory independent samples t-test ($t(76) = 2.079$, $p = .041$, d = .048) revealed that parents evaluated girls' capacity (M = 1.95, SD = 1.58) to make treatment decisions higher than boys' capacity (M = 2.75, SD = 1.43).

Table 4. GLMM of involvement of the child in decision-making (n=137)

	B	SE	t	P	Odds Ratio	95% CI for Odds Ratio Lower	Upper
Intercept	-4.406	1.531	-2.878	.005			
Age of Patient	.498	.058	8.569	.000	1.646	1.467	1.846
Female gender of Patient	1.296	.610	2.129	.035	3.656	1.096	12.193
Physician's Experience	-.602	.415	-1.450	.166	.548	.228	1.317
Prognosis of Disease	-.301	.243	-1.235	.219	.740	.457	1.198

Note. The dependent variable in this analysis is the involvement of the child so that 0 = no and 1 = yes. Results were adjusted for physician and centre clustering.

Discussion

By providing findings on children's actual involvement in decision-making, on parents' and physicians' evaluations of children's capacity to understand disease-related information and make treatment-related decisions, and on parents' roles in shared decision-making, this study presents new data contributing to the limited literature to date in shared decision-making in paediatric oncology, particularly in the Swiss paediatric oncology setting. The findings suggest appropriate and feasible ways to facilitate shared decision-making in paediatric oncology for all stakeholders. The study is unique as it highlights the dyad perspective on the same case.

Results from our dyad perspective first highlight that in comparison to physicians, parents rated the amount of information (on diagnosis, prognosis etc.) that they received as less satisfactory. Since studies have shown that most parents want to be informed honestly and frequently with respect to poor prognosis, this deficit in communication is likely to reduce parental satisfaction with decision-making[6,19,23,24]. For example, one study reported that the main reason for conflicts between physicians and parents was the latter's overly optimistic assessment of their child's prognosis[25]. In addition, parents perceived the fate of their children (i.e. treatment duration, suffering) as worse than how physicians perceived it. They thus felt that their children were suffering more and that the treatment seemed to be a long-lasting process. This divergence in the perception of information received could be because physicians avoided full disclosure to maintain hope. Although hope is a strong emotional motive, it may not produce the desired outcome in light of the value placed by the family on proper and adequate information in such situations[6,18,26]. On the contrary, full disclosure of prognosis is not only recommended by international guidelines[27] but can promote parental hope and peace of mind[19]. Other explanations for this difference are that information was not

sufficiently tailored to the parents' need, due to ineffective consent documents as well as difficulties associated with understanding complex information in a stressful situation with limited time[28]. There is thus a need to assess whether information provided is actually understood by the family[17] and a need for mechanisms to ensure clear communication between the healthcare providers and the family[29].

Second, parents held a more positive view of children's capacities as they rated the child's capacity to understand diagnosis and prognosis information higher than the physicians. This could be because they deemed their children more capable, perceived inclusion as being helpful, or were simply hopeful. Parent's more positive view raises the question whether physicians underestimate children's capacities or parents overestimate their children's abilities or whether the view of parents and physicians depend on factors not related to the child (e.g. the time when information was received, educational level of the parent, gender). Exploring the reasons behind parental and professional assessment of child's capacity is a fruitful area of investigation that is lagging presently[29].

Third, as expected our study findings point out that the likelihood of children's involvement in decision-making increases with age. While Hinds concluded that children between ten and twenty years of age are capable of participating in end-of-life decisions, in our sample only 69 per cent of this age group were involved, even though decisions considered in our study were not of this type and could be seen as being less cumbersome[26]. The qualitative findings from this project reveal that children and adolescents valued being involved in their treatment decisions[20,23]. Therefore, stronger involvement of children in light of their increasing age is recommendable for two reasons: age is highly correlated with the development of a child and involving children is internationally recommended[4,27,30]. Furthermore, guidelines highlight that children's level of understanding is often underestimated and that adolescents are aware of failed treatments[31,32]. Besides guidelines' recommendations and physicians' facilitation of

children's involvement in decision-making, parents have the responsibility to make their children's voices heard. However, this parental ability can be limited, for example, by the burden of coping with their child's disease[15] and exclusion of children from medical discussions because they wish to protect their child[33]. Related to inclusion of a paediatric patient, an interesting finding of our study is that girls were more likely to be involved even when there was neither age nor prognosis difference between boys and girls. An explanation from our exploratory analysis is that participating parents considered girls more capable of making treatment decisions than boys. Future research should carefully examine this finding.

Finally, similar to results from a study carried out in the United States, our study found that only 64 per cent of the parents held their preferred role, with 28 per cent holding a less active role and 8 per cent a more active role[5]. It should be noted that there was no difference between parents' and physicians' evaluation of the parents' preferred role in decision-making. That means that participating physicians in our sample perceived the parental preferences correctly but the realization of preferred roles was hindered. This is concerning since a study pointed out that holding a less active role was associated with lower evaluation of communication quality[5]. One reason for parents' less active roles could be that physicians were critical of the parental roles, namely parents holding too much decisional authority, and therefore restricted parents' participation[7]. In the face of their child's disease, parents often want to gather further expert opinions[28], and it could be that parents did not receive enough time to make a decision in light of the time constraints in clinical practice[34]. It is important to take parental preferences into account and to conduct research on decision-making because this can influence practice in paediatric oncology[35]. Thus, barriers that hinder shared decision-making and individual-level factors that affect such processes need further evaluation to close this gap between perceived and current parental roles.

Limitations

The limitations of this study include the different time range during which data was collected in the eight participating centres. One centre refused participation, but we do not believe that parents and physicians in that centre would have provided a significantly different response. Second, physicians carried out survey dissemination to the families. We can neither ascertain the number of families to whom the study was explained and study materials distributed nor the number of families who refused to participate. The response rate calculated in the methods section is limited to the number of surveys completed by the physicians which composed our known denominator. Third, 80 per cent of the participating parents were mothers. Since mothers are more likely to carry out the main responsibility for their child during these situations, it is a legitimate over-representation. Fourth, from the 151 children, the dyad-perspective was captured for only 78. Correspondingly, for 48 per cent of the children, only one perspective was available, and thus comparative analysis could not be performed for all children cases. However, the number of dyad-perspectives is sufficient to derive statements about differences between physicians and parents. Finally, as our aim was to gather information about children who had cancer, we did not differentiate their disease trajectory. Therefore, this information was not gathered in our survey, and there could be an effect on the results of the child's point along the disease trajectory. Given that participating parent and the physician completed their surveys on the same child (dyad-perspective) within a few weeks, it is not very likely that the point along the disease trajectory differed significantly within a dyad.

Conclusion

Our study provides both valuable insights into the decision-making of physicians and parents, and information to improve the decision-making process. It reveals the need for healthcare

providers to ensure that information provision is clear and correctly understood by the family. They should not take for granted that the information they relate to the family is perceived the way it is intended. That a girl patient is more likely to be involved in decision-making than a boy patient of the same age cautions both physician and parents to evaluate their perception of a child's capacity so that a capable male child is not denied participation. Additionally, our results note that physicians fail to ensure the preferred role of the parents. Measures to ensure that parents are enabled to enact their preferred roles in decision-making will be valuable to ensure good communication and the family's satisfaction with healthcare. Finally, our findings can be applied beyond paediatric oncology to the general aim of facilitating the optimal participation of parents and paediatric patients in shared decision-making.

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Chapter 3

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Palliative Care in Swiss Pediatric Oncology Settings: A Retrospective Analysis of Medical Records

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Abstract

Purpose: This study examined the provision of palliative care and related decision-making in Swiss pediatric oncology settings. The aim was to determine if and when children who died from cancer received palliative care, whether there were differences by cancer diagnosis, and inclusion of children in decision-making regarding palliative care.

Methods: Using a standardized data extraction form, a retrospective review of medical records of deceased pediatric patients was conducted. The form captured information on demographics, diagnosis, relapse(s), treatments, decision-making during palliative care, and circumstances surrounding a child's death.

Results: For 170 patients, there was information on whether the child received palliative care. Among those, 38 cases (22%) did not receive palliative care. For 16 patients, palliative care began at diagnosis. The mean duration of palliative care was 145 days (Mdn = 89.5, SD = 183.4). Decision to begin palliative care was discussed solely with parent(s) in 60.9% of the cases. In 39.1% the child was involved. These children were 13.6 years of age (SD = 4.6), whereas those not included were 7.16 years old (SD = 3.9). Leukemia patients were less likely to receive palliative care than the overall sample and patients with CNS neoplasms received palliative care for a longer time than other patients.

Conclusions: There are still high numbers of late or non-referrals and even children older than 12 years where not involved in decision-making regarding palliative care. These results do not align with international organizational guidelines which recommend that palliative care should begin at diagnosis.

Introduction

Since the early 2000's, pediatric palliative care guidelines have carefully distinguished palliative and hospice care, and ultimately recommended an integrative model for concurrent administration of curative treatment and palliative care[1-4]. This integrated model starts at diagnosis and continues throughout the illness trajectory, irrespective of the outcome[5,6]. It focuses not only on symptom management of the child, but also on the social, psychological, and spiritual well-being of both the child and the family[1,5]. However, palliative care (PC) for children is frequently deemed infeasible within the clinical practice for many reasons, including disagreement with the definition of PC as psychological and social support may already be part of the curative treatment[1]. It thus appears that the traditional understanding of PC, namely a dualistic model of curative and palliative, predominates in clinical care[7-9], and, accordingly, a chasm between guidelines' recommendations and actual clinical practice exists.

Although several studies highlight general improvement in the early implementation of PC[10,11], PC still does not always begin at the time of diagnosis of a life-threatening disease as recommended by existing guidelines. For instance, a Canadian study found that pediatric oncology patients did not receive PC at diagnosis, but most were referred during the course of their illness[9]. A survey of pediatricians in the U.S. concluded that children with life-limiting illness are referred to PC late, that is, at the end of their illness trajectory when no other curative options exist[7]. One study revealed an average time of 461 days after being diagnosed with cancer for pediatric patients to be referred to PC[9]. Similarly, a nationwide retrospective medical records review of pediatric oncology in Sweden found that the transition to non-curative care took place between the last day of life to over four years before death, with a median of 60 days[8]. The large range was attributed to varying types of cancer, as children

with leukemia were treated curatively until very close to death, while those with brain and solid tumors received PC earlier. An Australian study reported an average duration of 69.4 days of PC provision[12]. Lastly, a U.S. study found that the time between PC consultation and death ranged from 1 to 96 days (median 18 days)[13]. Reasons for late referrals are many: misconception of PC as not belonging to cure-oriented therapy[14], thus referring to PC only when no curative treatment exists; an uncertain prognosis compounded with families' refusal to acknowledge the incurable condition[15] and fear that the family may feel abandoned by primary caregiver if PC is discussed[16]; and physicians' difficulties with objectivity and uneasiness in diminishing hope[14].

Closely related to the issue of provision of palliative care for children is the question of their inclusion in such decisions. In Switzerland, there is no age at which children become legally competent to make decisions (Art 16. Swiss Civil Code). In the literature and in international guidelines, children's participation is recommended along with provision of information adapted to their personality, cognitive ability, maturity, and age[2-5,17]. Coyne and colleagues[17] found that healthcare professionals strongly supported a transparent approach to information sharing. However, parents wished to protect their children from burdensome information to maintain hope, and children trusted their parents to act as their advocates[17,18]. Several other studies underline the involvement of the patient and his or her parent in palliative cases[19-22].

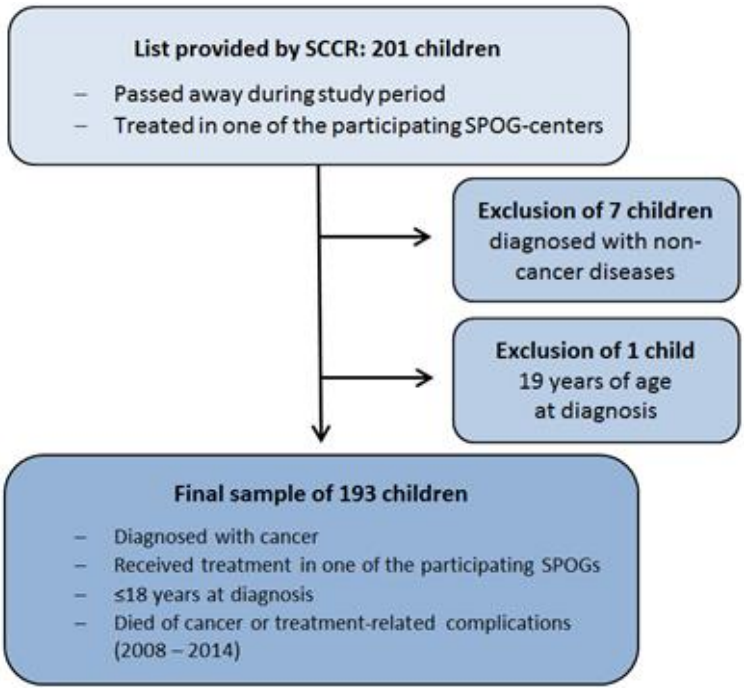
In sum, it is evident that a sizable fraction of children and their families still do not benefit from holistic and quality of life enhancing PC at early stages of the illness and data concerning children's inclusion in decision-making (DM) regarding PC is lacking. This study examined provision of PC in Swiss Pediatric Oncology Group (SPOG) settings to determine if and when children who died from cancer received PC, whether there were differences by cancer diagnosis, and inclusion of children in their DM process. We chose to raise these questions in the Swiss settings in response to our previous nation-wide project on treatment DM and inclusion of children in these decisions[20,22-24]. During the course of our interviews in

the previous project with children, their parents, and corresponding physicians as well as survey of parents and physicians, we were unable to obtain data on palliative cases mostly due to hesitancy to discuss this issue with patients and their families. Therefore, we choose to exclusively examine PC in this setting using a retrospective study design.

Methods

Seven of the nine SPOG-centers in Switzerland participated in this study because they had collaborated with the researchers on the previous project. Using a standardized data extraction form, a retrospective review of medical records of deceased pediatric patients was conducted. All responsible ethics committees approved this study. The list of children who comprised our study cohort was provided by the Swiss Childhood Cancer Registry (SCCR). Please see figure 1 for inclusion and exclusion criteria.

Figure 1. Inclusion and exclusion criteria



Data Collection

The data extraction form captured the following information: a) demographics, b) type and date of diagnosis, as well as number and dates of relapse(s), c) type of treatment(s), d) DM during PC, and e) circumstances surrounding the death of the child. These items were developed based on the research team's knowledge in the field and discussions with

collaborating physicians. Not all information was available in the medical records for all cases. In such cases, the researchers had the opportunity to consult the collaborating physicians. However, several missing values remained, which caused different sample sizes for some analyses. For example, the exact date when PC was initiated was not always available and consequently the duration of PC could not be calculated for these cases. The medical records were read carefully by four researchers who extracted the data. To ensure consistency of data collection, the first five extractions were discussed among the researchers; these discussions continued throughout the data collection period. Data was collected on-site from July 2015 to July 2016 and to ensure anonymity, an alphanumeric code was created for each child based on a predefined algorithm.

Data Analyses

All extracted data were entered into SPSS.22 by a research assistant and verified by another researcher for accuracy. Statistical analyses were performed using SPSS.22 (SPSS Inc, Chicago, IL). For analyses described below, reported *P* values are 2-sided and statistical significance level was set at $P < .05$. The variable of interest, PC, was identified either by an explicit reference to "palliative care" or when the records' content on treatment implied that PC was started, for example: curative treatment was stopped and quality of life was envisaged via best supportive care. The variable, "Provision of PC" indicates that children received PC (as per our interpretation of the medical records) characterized by a focus on quality of life and a supportive intent as compared to previous curative treatment with a focus on life prolongation and a restorative intent. The variable, "transition to PC" marks the point where it was clear from the medical records that curative treatment was no longer viable and thus, a PC was chosen.

After performing descriptive analyses, we divided our sample into three diagnosis-based subgroups (leukemia, CNS neoplasms, and other diagnoses) which were compared using

analysis of variance and Chi-square test of independence. Besides the quantitative analysis, content analysis was used in order to qualitatively analyze the discussions between medical professionals and the family[25].

Results

The average age of the sample (N=193) at diagnosis was 7.2 years and 55.4% were male (table 1). All 12 diagnosis groups of International Classification of Childhood Cancer 3 (ICCC3) were represented in our sample: CNS neoplasms (III) 34.2%, Leukemias (I) 27.5%, Neuroblastoma & other peripheral nervous cell tumours (IV) 11.9%, Malignant bone tumours (VIII) 8.3%, Lymphomas & reticuloendothelial neoplasms (II) 6.2%, Soft tissue & other extraosseous sarcomas (IX) 6.2%, and the remaining six formed 5.7%.

Table 1. Demographics of children by SPOG-center (N=193)

Center	Mean age at diagnosis (Mdn, SD)	Mean age at palliative care begin [b] (Mdn, SD)	Sex (male)
Center 1	7.5 (7.0, 5.0)	9.6 (8.0, 4.8)	68.4%
Center 2	6.8 (6.0, 5.2)	7.8 (7.0, 5.0)	56.6%
Center 3	8.2 (8.0, 5.3)	11.5 (13.0, 4.7)	63.6%
Center 4	6.5 (6.0, 3.5)	9.6 (9.0, 4.1)	35.3%
Center 5	9.3 (10.0, 4.3)	11.55 (13.0, 4.5)	58.8%
Center 6	6.0[c]	12.7 [c]	100%
Center 7	6.8 (6.0, 5.5)	10.0 (10.0, 5.4)	46.1%
Total	7.2 (6.0; 5.1)	9.5 (9.0, 5.1)	55.4%

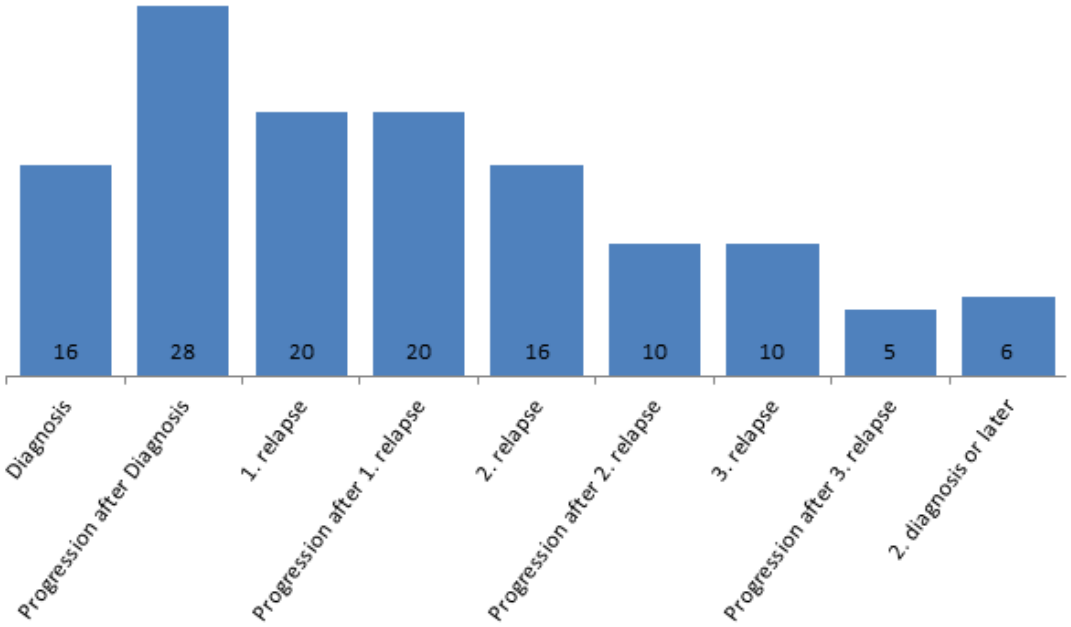
[a] In order to preserve anonymity we do not provide absolute numbers for each center; [b] palliative care time variable data available for 130 cases; [c] Due to the small number of children median and standard deviation were not reported.

When Did Palliative Care Begin?

For 170 patients, data was available on whether a child received PC. Of these, 77.7% (n=132) received PC and 22.3% (n=38) did not with the rationale being: in five cases, PC was discussed, but the child died before it could be started; in 25 cases, the child died before PC could be discussed; in four cases, the family opted for continuation of curative treatment even after physicians recommended PC; and in one case, parents refused both further curative treatment and PC. For three cases, there was no rationale documented in the medical records.

Of the 132 cases who received PC, information on when PC was started was available in all but one case. We found that PC began at diagnosis in 16 cases (12.1%) because of poor prognosis, and at progression after diagnosis in 28 cases (21.2%). In the remaining 88 cases, PC started at first relapse or later (66.7%; figure 2); for six children PC started after a second cancer diagnosis.

Figure 2. When did palliative care begin



Duration of Palliative Care

Duration of PC was computed using the start date of PC and time of death, which were available in 104 out of the 132 cases (79%). Of these children, 11.5% received PC equal to or less than one week, 19.3% between one week and one month, 21.1% between one month and three months, 22.1% between three and six months, and 26% more than six months. The mean duration of PC was almost five months, or 146.6 days (*Mdn* = 89.5, *SD* = 182.7, range: 2 - 1111 days). For six children (5.8%), PC began the day before their death.

Decision-Making Regarding Palliative Care

Information on who was present at palliative care DM was found for 115 cases and on children's involvement for 113 cases. Decision to begin PC was discussed solely with both parents or with one parent in 60.9% of the cases (70 out of 115), with both or one parent and the child in 34.8% of the cases (40 out of 115), and first with the parents and afterwards with the child in 4.3% of the cases (5 out of 115). Children who were included in DM were on average 13.6 years of age (*SD* = 4.6), whereas children that were not included were on average 7.16 years of age (*SD* = 3.9; table 2).

Table 2. Involvement of children (n =113)

Age	Not involved (n=70)	Involved (n=43)
0	2	0
1	1	0
2	4	0
3	4	1
4	6	0
5	12	2
6	7	2
7	6	1
8	4	1
9	5	2
10	5	1
11	2	2
12	1	0
13	6	4
14	1	6
15	1	5
16	1	4
17	2	5
18	0	6
19	0	1

Although the participating SPOG-centers used different names (e.g. ethical committee, dialogue ethic), all centers generally employed discussion of ethically relevant and challenging issues, if indicated. Information on whether ethics relevant discussion took place was available for 175 cases. In 61 cases an ethical discussion involving different professions

was conducted (excluding family members), and for the remaining 114 cases, no information on ethical discussions was found.

Using the written statements in the medical records where information was given on discussions between medical professionals and family members, we categorized them into types of decisions and if children were included in these decisions (table 3). This resulted in eight types of decisions, ranging from refusing a discussion to shared DM. Most of the decisions were described as a one-time act.

Table 3. Results from the content analysis – decision making regarding PC

Type of decisions	**In which only parents were involved**	**In which child were involved**
Parents refuse discussion (n=1)	"(The transition) was discussed among the medical staff, parents refused to discuss it."	-
Parents want physician to make the decision (n=1)	"She (the mother) expresses the desire that in the case of further progression of the disease, she does not want to make that decision (to continue or stop curative treatment). She wants the physicians to make that choice..."	-
Withheld information from the child (n=5; children's ages; 9, 16, 11, 7, 10)	"The parents asked the physicians not to explicitly inform the child about the situation, and their request was respected."	-
Patient is aware of situation (n=6)	-	"Patient was also aware of the bad prognosis and was living his life in the present, without making any project in the long term."
Family express wish for palliative care (n=11)	(n=5) "In light of these facts (chance of survival or cure very low) parents themselves suggested the shift towards best quality of life, symptom control and palliation (…), physicians agreed that this makes sense."	(n=6) "Family decided to receive palliative therapy at home, wish on the part of patient and family."
Agreement with physician's decision (n=16)	(n=10) "After diagnosing third relapse: discussion with parents; given the progression of a third relapse the parents were informed that there is no curative option available; Parents agreed to palliative care."	(n=6) "Palliative treatment was recommended to parents and patient; Parents and patient agreed"

Parents informed about decision made by the physicians (n=30)	(n=22) "Because of huge suffering and the low likelihood to cure the patient: palliative therapy in order to achieve freedom from pain without loss of vigilance; parents were informed about the consensus at the same day."	(n=8) "They (parents and patient) were informed "openly and in detail" about infaust prognosis in case of progression under chemotherapy; from now on: palliative treatment with focus on symptom management and QoL"
Shared decision making (n=48)	(n=28) "After several discussions, the parents and the physicians decided to stop the chemotherapy treatment that had been initially intended and to begin the comfort care."	(n=20) "The child and his family were told that the chances of cure were almost inexistent … After several days, the child and his family chose to follow the combined treatment (radiotherapy and chemotherapy through bone, with palliative goal)."

Diagnostic groups and palliative care

Three diagnosis-based subgroups described in the literature on paediatric cancer patients[8,26,27] were created: (1) leukaemia patients, (2) CNS neoplasms patients, and (3) patients with other diagnoses. These groups were compared with respect to whether a child received PC. The relation between *whether PC was received* and the diagnostic groups was significant, X^2 = 30.9 (2, N = 170), p = .000, indicating that diagnostic group has an impact on whether a child receives PC. Post hoc tests (Bonferroni correction was applied) illustrated that patients who died from leukemia were less likely to receive PC (p = .000), and patients with other diagnoses were more likely to receive PC (p = .008) than the overall sample. There was no significant result for patients with CNS neoplasms.

Furthermore, analysis of variance between the three diagnostic groups was conducted for further variables related to PC, revealing that there were significant effects of diagnostic groups on both initiation and duration of PC (table 4).

Table 4. Summary of ANOVAs with respect to palliative care

Variable	*M* (N)			df	*F*	ω^2	*p*
	Leukemia	CNS	Others				
Begin of PC[b] *	5.22 (23)	3.00 (47)	4.18 (61)	2	9.290	.11	.000*
Duration of PC in days[a] **	81 (22)	224 (38)	113 (44)	2	6.112	.09	.003**

Note. PC = palliative care; CNS = CNS neoplasms; Begin of PC represents an ordinal variable that corresponds to the nine ordered categories shown in figure 2. [a] Welch ANOVA (an adjusted omega squared and Games-Howell post-hoc test were used); [b] One-way ANOVA (Tukey post-hoc test was used).

∗ PC started later for both leukemia patients (*M* = 5.2, *SD* = 1.6) and patients with other diagnoses (*M* = 4.2, *SD* = 2.2) as compared to patients with CNS neoplasms (*M* = 3.0, *SD* = 2.2). The former two did not differ significantly. ∗∗ Patients with CNS neoplasms (*M* = 223.8, *SD* = 236.4) received PC for a longer time than both patients with leukemia (*M* = 81.4, *SD* = 138.6) and with other diagnoses (*M* = 112.5, *SD* = 117.6). The latter two did not differ significantly.

Discussion

The integrated model of PC recommends that it commence alongside curative treatment[5,6], thus ensuring that all children with life-threatening diseases benefit from this approach irrespective of prognosis. First, in our study sample, one out of eight children received PC at the time of diagnosis due to poor prognosis. For the remaining cases, there was a transition to PC with the realization that a curative intent would be ineffective. These results support the findings of Johnston and colleagues[9] that most patients received PC but these referrals seldom occurred within a month of diagnosis. Unlike other studies[10,11], we have no previous nationwide study to compare whether PC provision has improved in the country. However, recent investigations into PC in pediatric oncology do indicate that it is an important topic of national interest[20-22,28,29]. Second, 15% of our sample received PC for a week or less because PC was implemented very late or at the end of the illness trajectory[7]. Third, our results also indicate that the mean duration of PC of 145 days was longer than in analogous studies performed in Sweden[8], Australia[12], and the U.S.[13] The longer PC duration may indicate earlier integration of PC in our sample, but can also be attributed to methodological differences (e.g. different definitions of when PC started), cultural perceptions of when a condition cannot be cured anymore (e.g. prognosis smaller than 10% versus 5%), or the composition of the sample.

Because discussing PC is sometimes associated with loss of hope and abandonment by the healthcare provider[16], it might be a strong argument for physicians to not call some of their efforts towards relieving pain and supportive care as PC and to use a less distressing term. There may be hesitations to discuss PC by explicitly mentioning the term (and consequently not recording it as such) even when it occurred or the term was avoided because supportive care and symptom management are generally considered parts of oncological treatment.

Given physicians' tendency to avoid the term and families' discomfort when PC is suggested[16], it might be advantageous to use a term that is less threatening in this context, such as best supportive care. It has to be noted that, if this term was used (or any other term), it must be unambiguously defined to avoid conceptual confusion. Further, it should be clearly explained to families to avoid any misunderstanding associated with the terminology and, consequently, to help them better accept the initiation of PC at diagnosis. However, studies have shown benefits of providing PC consultation and including a PC specialist[11,13], such as identifying the need of medication changes. Finally, irrespective of the terms used, this particular type of care has to be compassionate, individually tailored to the needs of the child and the family, and holistic embracing multiple domains of care such as physical, psychological, social, and spiritual care. Providing adequate PC also includes developmentally appropriate preparation for death.

Comparisons of diagnostic groups suggest that leukemia patients began PC later than CNS neoplasms patients and patients with other diagnoses did, their duration of PC was shorter than that of CNS neoplasms patients, and they were less likely to receive PC than the overall sample. Also, the PC duration of patients with CNS neoplasms was significantly longer than for patients with other diagnoses, and the latter were more likely to receive PC than the overall sample. These findings relate to the Swedish study[8] in which children with hematological malignancies received curative treatment closer to death and transitioned later than children suffering from brain tumors. One explanation could be the higher survival rate of children suffering from leukemia compared to children with other types of cancer[30-32]. Additionally, successful curative treatment protocols are available in leukemia relapses using different treatment modalities, such as conventional chemotherapy, high-dose chemotherapy, allogenic hematopoietic stem cell transplantation, and many different experimental drugs. Therefore, there may be greater hesitation by the healthcare provider to recommend PC at diagnosis, as well as higher expectations of maintaining hope in leukemia cases[14,16], whereas

for CNS neoplasms patients, comparably bad prognoses might prompt the physician to begin PC earlier.

Although we did not collect data on the quality of communication and information provided, it was evident that parents and, in some cases, children were involved in the DM process (either actively in shared DM or passively in "informative" DM) and information was thereby given to them. Given the information in the medical records, shared DM appeared to be the most frequently used approach to DM in our study. While this reflects only the physicians' perspectives and families may have perceived the DM process differently, this finding is in line with the calls for such a DM process in pediatric healthcare[17,33-35]. At the same time, contrary to the suggestion that clear and appropriate information provision is helpful for young patients[5,17,36], in five cases, it was explicitly stated that information was withheld from the patient per their parents' wishes. The WHO and pediatric association guidelines recommend inclusion of children in their DM[2-4], which was not the case for all children in our sample. Specific to Switzerland is the issue of capacity, as most children aged 14, and many aged 12 are considered competent[37,38] and therefore legally have the right to be included in their DM. With respect to DM and PC, Whitney and colleagues propose that in situations where cure is unlikely and PC services are the best option, the physician should take the lead in decision making[39]. That is, they should exercise decisional priority by identifying the single best course of action and should explain to the parents (and child) that there is no curative treatment left and thus, quality of life becomes the new focal point. However, decisional authority, that is the nondelegable right of the parents to decide, remains with the parents and, as the child matures, decisions should be increasingly shared with the child.

This study has several limitations. The findings are limited to the quality of information available in the medical records. Not all information was available, resulting in missing values for several variables, which, in turn, resulted in different sample sizes for some of the analyses. It is also possible that the extracting researchers could not find the information in

the medical records or that the information they were looking for, e.g. PC, was a term that was not used by the physicians unless it was absolutely clear that the case was end-of-life. However, sample sizes were sufficiently big to apply inferential statistics. Furthermore, the quality of information can vary across centers and individual health staff members. As the medical records were written in French, German, or Italian and translated into English by our team, we cannot exclude linguistic differences which could affect interpretation of the data. During the time covered by our analysis (2008 – 2014), the pediatric team involved in PC may have changed, resulting in variations in the information documented in medical records over time. Finally, the views of the families and children themselves are not directly represented, as the information in the records is solely noted by medical staff and is their impression of the communication between physicians and family. It is necessary to understand the views of family members as well to obtain a better picture of how provision of PC and DM surrounding it took place. Therefore, further studies are necessary to fully capture the intricacies of PC for children with cancer and inclusion of children in such delicate DM. Limited evidence exists on this issue in Switzerland, and considering the limitations of a retrospective study that used medical records, prospective studies are needed to further strengthen research in this field. In addition to more research, the medical team and the family may benefit from the introduction of a standardized form for the recording PC discussions and decision-making. Such standardization may also improve communication within the team as well as with the family and, thereby, facilitate shared DM.

In conclusion, the data on PC in the Swiss pediatric oncology settings presented in this study underline that only a very small proportion of the children received PC at diagnosis and for most children palliative care began late in the illness trajectory. These results on the timing of PC do not align with international organizational guidelines which recommend an integrated approach. However, integrated PC has been shown to be beneficial for children[10,11,40] and therefore, must be envisaged. Additionally, since it is not only the right of children to be

involved in decisions that affect them, but also a need of a child[23], the possibility of including them in DM must be considered. Furthermore, our findings from the medical records could help discuss the usage of the terms best supportive care and PC, which as shown from our data collection, seems to be a term mostly understood as the opposite of curative care. Finally, reasons for not including children older than 12 years need to be further examined, especially for the Swiss context in which capacity of judgement is not strictly defined based on age of maturity and minor patients between 12 and 18 years are mostly expected to be capable of judgement.

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Chapter 4

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Burden of Treatment in the Face of Childhood Cancer: A Quantitative Study Using Medical Records of Deceased Children

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Abstract

Lived experiences of childhood cancer patients and their families have been described as interrupted and as a loss of normal life. Apart from symptoms due to the cancer disease, families continuously experience burden of treatment. Since coping capacities are unique to each individual, we captured variables that offer objective measures of treatment burden, with a particular focus on the disruptive effects of treatment on families' lives.

Our sample was comprised by 193 children that died of cancer. Medical records were extracted retrospectively. Quantitative data was statistically analysed with respect to variables related to treatment burden.

Deceased children with cancer and their families faced a significant burden of treatment. Results revealed that deceased leukemia patients had a higher number of inpatient stays, spent more time in the hospital both during their illness and during the last month of their life, and were more likely to die in the hospital when compared to deceased patients with CNS neoplasms and with other diagnoses.

Our findings highlight the disruptive effects of treatment that are likely to have a great impact on families' daily life, that go beyond exclusively focusing on side effects, and that needs to be taken into account by the treating staff.

Introduction

Worldwide, approximately 200.000 children are diagnosed with cancer each year, among them 50.000 from high-income countries with a survival rate around 80%[1]. Living with a life-threatening disease like cancer represents a burdensome and difficult time for both the pediatric patient and the family[2].

The Cancer Experience for Children and their Families

A cancer diagnosis marks an abrupt break in life. Lived experiences of children with cancer and their families have been described as an interrupted one with limited opportunities to engage in leisure activities and difficulties in sustaining existing friendships[3-5]. Cancer diagnosis of a child is often described as a loss of normal family life[2,6,7]. Moreover, children diagnosed with cancer experience being isolated from friends and family members [8,9], which may have negative consequences on their well-being[10-12].

The interruptive effect of cancer on the everyday life of the child as well as his or her family can be attributed to the treatment procedures, their corresponding appointments, and waiting times[4]. In a qualitative study, children evaluated the latter as an even more significant stressor than the treatment itself because during waiting times they feel alone and anxious about what will follow[13]. Other studies show that children experience treatment procedures as highly stressful and their situation as one of having no choice but to undergo treatment[5,14]. In light of the changes that a cancer diagnosis brings, families seek to maintain as much of their former life as possible[4,15]. It is thus recommended that the inpatient stays of a child should be limited to what is necessary by drawing on outpatient visits and provision of care at home[16,17]. In support of this recommendation, a study found that when chemotherapy treatment was provided at home versus in the hospital, children had more opportunities to engage in normal

activities, parents were financially less burdened and saved time, and families' overall life was minimally disrupted[18].

In cases where children died due to cancer, studies found that most parents not only plan for their child's death, but they also favor their home as the location for death as well as for end-of-life care[19-21]. Besides the explicit parental wish that the child dies at home, various other determinants of place of death for children dying due to cancer have been identified, for example, diagnosis, length of last hospital admission, ethnicity, gender, or a child's age[22-24]. However, a systematic literature review found no compelling evidence that families opt home as the place of death[25].

The Burden of (Cancer) Treatment

Apart from symptoms due to the underlying cancer disease, families continuously experience burden due to the treatment(s) that the child must undergo. Burden of treatment is different from burden of illness. The latter expression rather refers to the physical and psychological effects caused by the illness[26]. The former points not only to the treatment side effects but to the burden which is associated with the treatment of an illness[27]; and "encompasses (among other things) the disruptive effects that treatment has on working lives (for example, having to repeatedly go to clinics for tests) and on social lives (for example, having to curtail activities because of treatment side effects)"[28]. Moreover, according to a literature review on the different conceptualizations of burden of treatment, the latter is in its nature dynamic, comprises both subjective and objective dimensions and distinguishes between antecedents and consequences of burden[27]. Finally, burden of treatment theory refers to the importance of relational networks when facing treatment because these "collective agents" are able to provide more resources than the individual patient[29].

Even though burden of treatment is unavoidable, it can function as an indicator for the quality of care provided or managed by the health care team[28]. A better understanding of burden of treatment may facilitate an improved understanding of the family's situation and preferences which, in turn, is a prerequisite for a minimal disruptive medicine that reduces patients', families', and caregivers' overall burden[26].

Study Purpose

Although it is understood that childhood cancer entails high burden of treatment as well as high burden of illness, there is a lack of studies that allow complete understanding of burden of treatment for children with cancer and their families. Our empirical work seeks to assess different indicators of burden of treatment for children who died of cancer through a study carried out in Swiss Pediatric Oncology Group (SPOG) centers. Since coping capacities are unique to each individual, we captured variables that offer objective measures of burden of treatment, namely time periods and count information, with a particular focus on the disruptive effects of treatment on families' lives. Examples of such variables include the number and duration of a child's inpatient stays which, among others, cause the above mentioned disruptive effects on families' daily lives. We retrospectively extracted these variables from medical records of children who died due to childhood cancer. This paper addresses two research questions: (a) a data based description of what the actual burden of treatment for childhood cancer patients who died of the illness and their families is in Switzerland, and (b) whether burden of treatment differs by different types of cancer.

Methods

Study Design

In Switzerland, there are nine SPOG-centers. We collected data from seven of them based on prior research collaboration. A retrospective review of medical records of pediatric oncology patients who died between 1 January 2008 and 31 December 2014 was conducted. Ethical approval was obtained from the responsible ethics committees in Switzerland.

Study Population

We extracted information from medical records of children who met the following inclusion criteria: the child (a) was diagnosed with cancer and received treatment in one of the participating SPOG centers, (b) was less than or equal to 18 years of age at diagnosis, and (c) died of cancer or treatment-related complications. We obtained information on which children died at each participating SPOG center from the Swiss Childhood Cancer Registry (SCCR). This information enabled us to exclusively search for children that met the inclusion criteria (note: SCCR includes information on both living and deceased pediatric oncology cases and reports data on adolescents up to 20 years of age). This study confined their analysis to children who died due to childhood cancer in response to our previous project on children's inclusion in treatment-related decision-making in which we were unable to recruit palliative cases due to hesitancy to bring up this issue within discussions with families[30-33]. Therefore, to examine decision-making regarding palliative care we decided to exclusively analyze medical records of deceased children.

Data Extraction Form

A data extraction form was designed to gather data on the following aspects from the medical records: a) demographics, b) diagnosis and relapse(s), c) treatments received, d) decision-making in the course of treatment, and e) death of the child. Items were developed from the research team's knowledge in the field and based on discussions with collaborating physicians. The extraction form consisted of items with categorical responses (e.g. center, sex, diagnosis), continuous variables (e.g. age, number of inpatient stays, time being inpatient), and open-ended items (e.g. Please note any information about the transition from curative to palliative phase). The data extraction form was used as a digital version (i.e. Microsoft Word document).

Data Collection

Before starting data collection, we contacted the collaborating physician of the respective SPOG-center in order to be referred to the SPOG-center's data manager who is responsible for research-related data requests. The data manager created access to the archive, organized working spaces, and remained at our disposal for further questions. Most medical records were available in paper form. In cases of already digitalized records, we extracted the relevant information from the digital records. Four research assistants gathered relevant data from the medical records using the extraction form. Data was collected center by center and on-site between July 2015 and July 2016. Researchers discussed the first five extractions aiming for standardization of extracting and continuously discussed their extractions, when needed. Each child was anonymized through a participant code at the time of extraction. This code was created based on a predefined algorithm.

Study Sample

Based on the list that was provided by SCCR 201 children died during the study period and were treated in one of the participating SPOG-centers. For seven cases, in consultation with collaborating oncologists the research team decided that the diagnosis was non-cancer and therefore excluded them, and one child was over the age of 18 years at diagnosis. After excluding these eight cases, a final sample of 193 cases remained.

Statistical Analyses

A research assistant entered all extracted records into SPSS.22 (SPSS Inc, Chicago, IL) and another randomly checked 15% of the data for correctness of data entry. Statistical analyses were performed using SPSS.22 (SPSS Inc, Chicago, IL). Since not every item of the extraction form was always contained in all medical records or could be found, sample sizes for some variables were less than the total number of data collected (N = 193). Information on palliative care and transition to palliative care are discussed in another paper. Related to the goal of this project, children's and families' burden of treatment was measured using the following seven variables: (a) number of inpatient stays, that is, the sum of all inpatient stays a child went through (n = 162); (b) total time being inpatient representing the durations of all inpatient stays (n=162); (c) duration of illness, which was defined as the time between diagnosis and death of the patient, thereby capturing the total time span a child had lived with illness (n = 185; This definition intends to capture both active treatment and in-remission time spans with the underlying rationale that even in periods when the child was in remission families' daily life was still somehow affected by the illness, for example by regular follow-up appointments, uncertainty because of possible relapses, or long-term effects on physical and psychological health.); (d) inpatient-proportion was computed using b and c (n = 154), for example, a patient was hospitalized 30 days and the entire duration of illness was 180

days, the percentage equals 16.7%; and (e) days being inpatient during the last month of a child's life, that is, burden through hospitalizations in the last month of their lives (n = 157). Finally, we analyzed (f) where a child died, namely at home or in the hospital (n = 161), and (g) who was present at death (n = 119).

In a first step, variables were analyzed descriptively. In a second step, we compared three diagnosis-based subgroups within our sample of deceased childhood cancer patients, namely leukemia patients, CNS neoplasms patients, and patients with other diagnoses employing analysis of variance and Chi-square test of independence. For the analyses, reported *P* values are 2-sided and statistical significance level was set at $P < .05$.

Results

Characteristics of the Sample

Fifty-five percent (107 of 193) of the deceased children were male. Mean age at diagnosis was 7.2 years, ranging from 0 to 17 years of age. Of the 12 categories (I-XII) of International Classification of Childhood Cancer 3 (ICCC), the most frequent diagnoses were as follows: CNS neoplasms (ICCC-III, 34.2%) and leukemia (ICCC-I, 27.5%). The other ten categories were also represented in our sample, and we collapsed the least six as "others" (table 1).

Table 1: Sample characteristics (N = 193)

Demographics	Mean (Mdn, SD) Percentage
Sex (male)	55.4%
Age at diagnosis	7.2 (6.0; 5.1)
Age at death (n = 179)	9.8 (9.0; 5.2)
Main diagnostic groups according to ICCC	Percentage
CNS neoplasms (III)	34.2%
Leukemias (I)	27.5%
Neuroblastoma & other peripheral nervous cell tumours (IV)	11.9%
Malignant bone tumours (VIII)	8.3%
Lymphomas & reticuloendothelial neoplasms (II)	6.2%
Soft tissue & other extraosseous sarcomas (IX)	6.2%
Others	5.7%

Note. ICCC = International Classification of Childhood Cancer 3

As expected, compared to the SCCR, a national dataset that includes all children diagnosed with cancer [34], due to the different mortality rates, leukemia was underrepresented in our sample of deceased children (27.5% vs 33.6%), whereas CNS neoplasms (34.2% vs 22.9%) and neuroblastomas were overrepresented (11.9% vs 6.1%). Patients' age was overall comparable to SCCR (in bracket): 0-4 years 37.8% (35.4%), 5-9 years 26.9% (21.3%), 10-14 years 24.4% (22.7%), and 15-18 years 10.9% (20.6%).

In most of the cases (77.3%; 140 out of 181 children) cancer treatment protocols were followed at diagnosis. In more than half of the cases (54.4%; 99 out of 182) other hospitals

were consulted for further support or advice. More than three quarter of the children received palliative care (77.6%, 132 out of 170, palliative care is defined here as opposed to curative care, i.e. palliative care means here a supportive care approach that was taken once it was agreed that the treatment goal was not anymore curative). This finding means that almost one quarter of the deceased children died during supposedly curative care and did not go through a phase where it was transparent that they were dying and during which they received only supportive palliative care (22.4%, 38 out of 170). Among them, 9 children died due to unexpected complications or side effects of the treatment.

Burden of Treatment

Our analysis focused on variables indicating deceased children's burden of treatment (table 2). On average, children from our sample had 13 inpatient stays during their illness course, which amounted to 117 days in the hospital. During the last month of their lives, the deceased children were hospitalized for an average of 10 days.

Table 2: Variables indicating burden of treatment

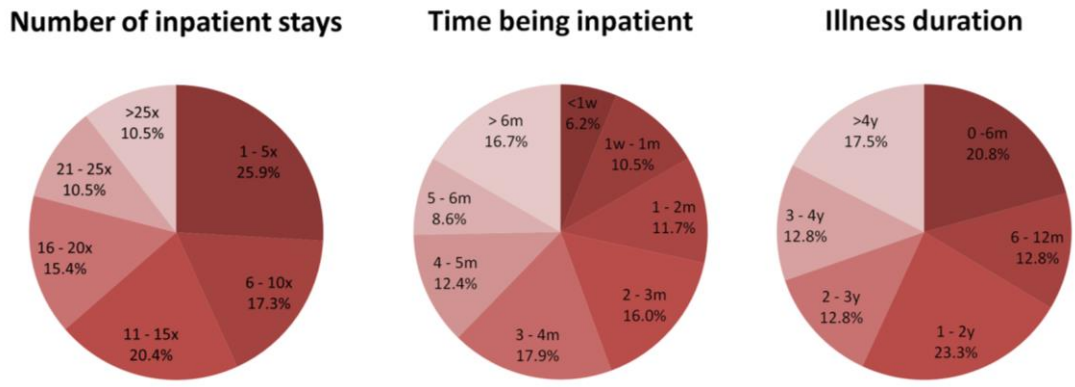
Variable	Median (M, SD) Percentage
Nr. of inpatient stays (n = 162)	12.0 (13.0, 9.2)
Days being inpatient (n= 162)	102.5 (117.3, 92.8)
Illness duration in months (n = 185)	19.0 (30.6, 34.2)
Inpatient days/ last month of life (n = 157)	5.0 (10.0, 10.8)
Inpatient-proportion in percent (n = 154)	15.0 (26.0, 24.8)
Child died at home (n = 161)	43.5%
Parents were present at death (n = 119)	96.6%

Overall, the majority of the children did not die at home: 55.9% died in the hospital (90 out of 161) and 0.6% (1 out of 161) in a supportive care center. Still, a relatively large minority of 43.5% of the children died at home (70 out of 161). Looking only at those children who received palliative care (and for whom we were able to gather information on location of

death), the numbers change as follows: 53.7% died at home (66 out of 123), 46.3% in the hospital (57 out of 123). At the time of death, in almost all of the cases parents were present except for 4 cases.

Figure 1 provides detailed information on burden of treatment, that is, number of inpatient days, time spent in inpatient care, and illness duration. First, deceased children's burden due to the number of inpatient stays ranges from 1 to 39 stays with almost one third of them having more than 16 inpatient stays. Additionally, one in ten children was admitted to the hospital more than 25 times during the entire illness period (figure 1). Second, concerning inpatient time, ranging from 2 to 581 days, one in six children lived in the hospital for 6 months or more (figure 1). Also two-fifths of the sample had between 3 and 6 months of inpatient stays. Third, the overall duration of illness ranged from 1 day to 5582 days (equaling around 183 months or 15 years, respectively), with more than one third of the children undergoing treatment for less than one year and almost one in five children for more than 4 years.

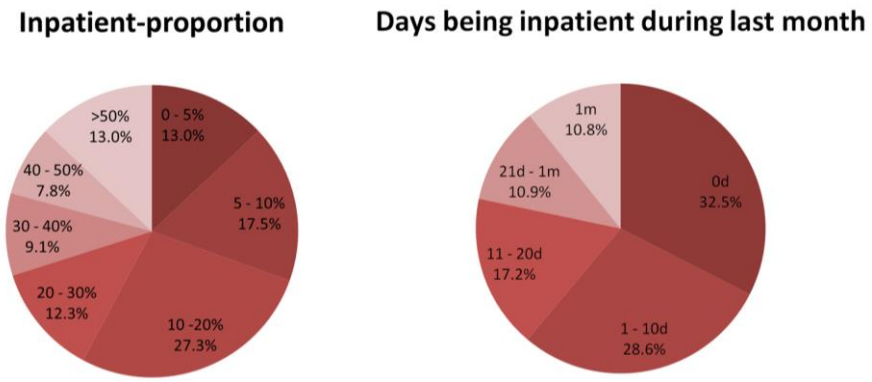
Figure 1: Inpatient stays and illness duration



Note. w = week; m = month; y = year.

To further understand the burden of treatment in our sample, we computed the percentage of time spent in the hospital during the entire duration of illness. The results show that almost one third of our deceased children sample spent less than 10% of their time during their entire illness duration in the hospital and more than one quarter between 10% and 20%. Almost one in eight children spent more than half of their illness duration in the hospital (figure 2). Furthermore, 29% of the children were hospitalized between one and ten days during the last month of their life, and one in ten children was hospitalized the entire last month of his or her life (figure 2). Moreover, an exploratory independent samples t-test revealed that children who died during curative treatment spent significantly more days in the hospital (n = 24, M = 16.8, SD = 11.7) than children who died during palliative care (n = 112, M = 8.2 days, SD = 9.7), t (134) = -3.779, p = 0.000.

Figure 2: Inpatient-proportion and days being inpatient during last month



Note. d = day; m = month.

Burden of Treatment: Comparisons among Diagnostic Groups

Based on the twelve diagnostic categories of ICCC of our sample, we reclassified them into those who died with: a) leukemias (27.5%), b) CNS neoplasms (34.2%), and c) other diagnoses (38.3%). Leukemia and CNS neoplasms were chosen to represent individual groups because they formed the two biggest groups in our sample and were already used in

studies on pediatric oncology [22,23,35]. Since summing up the remaining ten categories of ICCC resulted in a comparably big subgroup, they were grouped into the residual category "other diagnoses". Hereinafter, this diagnosis variable is referred to as "three diagnostic groups". Subsequently, analysis of variance (table 3) and Chi-Square test of independence were conducted for variables related to children's burden of treatment.

Table 3: Summary of ANOVAs

Variable	*M* (N)			df	*F*	ω^2	*p*
	Leukemia	CNS	Others				
Nr. of inpatient stays[b]	14.3 (44)	8.2 (50)	15.8 (68)	2	11.818	.12	.000
Days being inpatient[a]	155.8 (44)	85 (50)	116 (68)	2	7.461	.05	.001
Illness duration in days[b]	857.4 (50)	933 (64)	986 (71)	2	0.220	-	.802
Inpatient-proportion in percent[b]	36.7 (41)	21.9 (49)	22.3 (64)	2	5.510	.06	.005
Inpatient days/ last month of life [b]	15.8 (46)	6.6 (49)	8.4 (62)	2	11.035	.11	.000

Note. CNS = CNS neoplasms; df = degrees of freedom (between); *F* denotes the F statistic used with ANOVAs; ω^2 denotes the effect size omega squared, an adjusted omega squared was used for Welch ANOVA; *p* = p-value; Depending on homogeneity of variance, Welch ANOVA or One-way ANOVA was used. [a] Welch ANOVA; [b] One-way ANOVA.

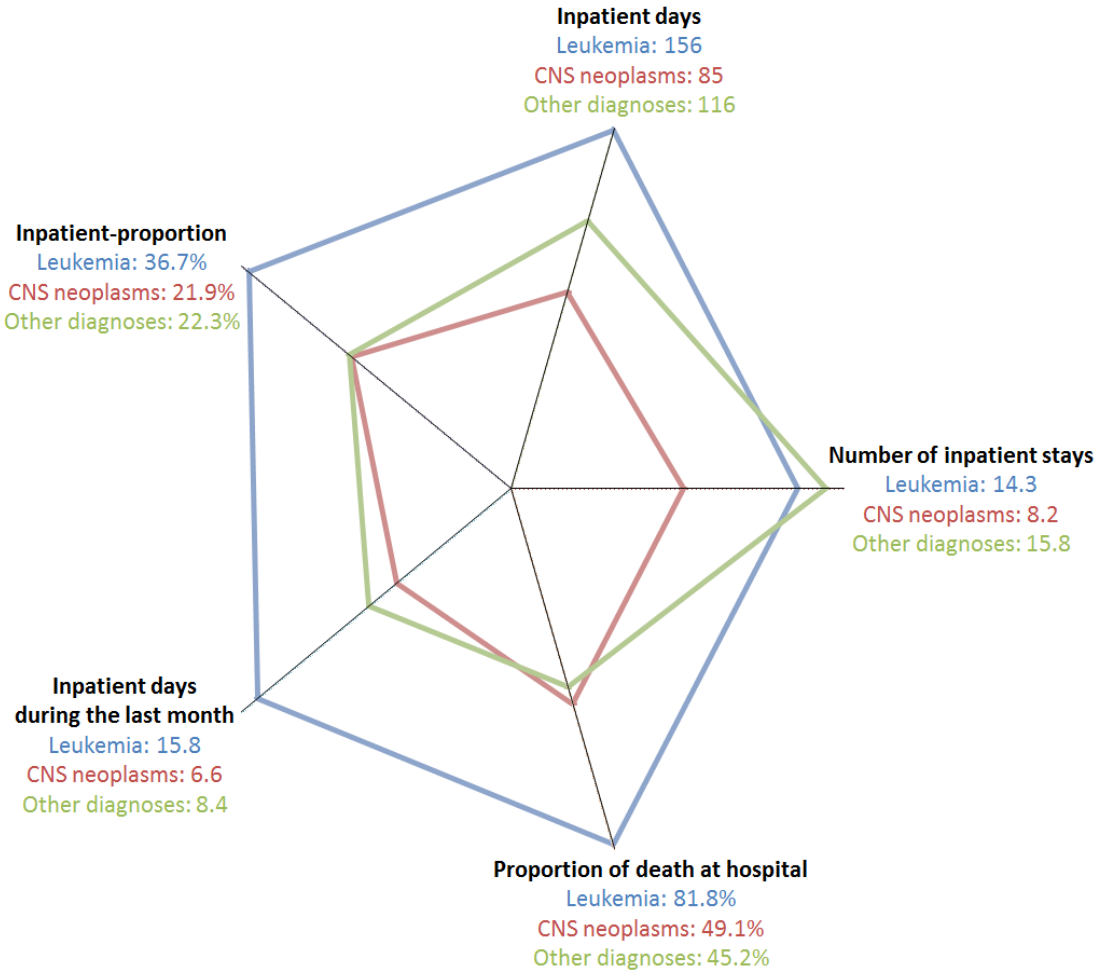
Analysis of variance revealed that there were significant effects of the diagnostic groups on the deceased children's number of inpatient stays, the days being inpatient, the inpatient-proportion, and inpatient days during the last month of life. Post hoc comparisons for the four significant results were conducted in order to see which diagnostic group or groups differed from one another.

First, with respect to the *number of inpatient stays*, a post hoc Tukey test showed that both leukemia patients (*M* = 14.3, *SD* = 9.0) and patients with other diagnoses (*M* = 15.8, *SD* = 9.0) had significantly more inpatient stays than those patients with CNS neoplasms (*M* = 8.16, *SD* = 7.9). There was no significant difference between leukemia patients and patients with other diagnoses (figure 3).

Second, for *days being inpatient*, a post hoc Games-Howell test revealed that leukemia patients (M = 155.8, SD = 122.0) had significantly more days being inpatient than patients with CNS neoplasms (M = 84.5, SD = 88.9). No other pairwise post hoc comparison revealed a significant difference (figure 3).

Third, concerning *inpatient-proportion*, a post hoc Tukey test showed that leukemia patients (M = 36.7%, SD = 26.4%) spent significantly more time of their overall illness duration in the hospital than both patients with CNS neoplasms (M = 21.9%, SD = 27.8%) and those with other diagnoses (M = 22.3%, SD = 19.0%). The latter two did not differ significantly (figure 3).

Figure 3. Comparisons among diagnostic groups



Fourth, regarding *days being inpatient during the last month of life*, a post hoc Tukey test showed that leukemia patients (M = 15.8, SD = 10.9) were hospitalized for more days than both patients with CNS neoplasms (M = 6.6, SD = 9.8) and the remaining with other diagnoses (M = 8.4, SD = 9.7). The latter two did not differ significantly (figure 3).

Finally, the relation between the *place of death* (at home or in the hospital) and the diagnostic groups was significant, X^2 (2, N = 160) = 15.9, p = .000, V = .32, indicating that the diagnostic group has an impact on whether a child dies at home or in the hospital. Post hoc tests (Bonferroni correction was applied) revealed that leukemia patients (81.8%) were more likely to die in the hospital (p = .000) than the total sample (56.5%). There was no significant result for patients with CNS neoplasms or for patients with other diagnoses (figure 3).

Discussion

Overall, assessing the treatment-related burden descriptively reveals that deceased children with cancer and their families face a significant number of inpatient stays and long hospitalizations. Undoubtedly, the many inpatient visits lead children to miss out on school and parents from work, changes in both familial and social relations, limited opportunities for leisure time activities, as well as a general physical and psychological strain. Such effects of childhood cancer are described as "rough spots"[36-38]. Since our sample was exclusively comprised of children who died due to cancer, they may have faced a higher burden of treatment. As a consequence, further research examining burden of treatment among children who survived is needed.

Deceased Leukemia Childhood Cancer Patients: The Higher Burden of Treatment

Specifically, deceased leukemia patients faced an almost double burden (number of inpatient stays and time being inpatient) compared to those patients with CNS neoplasms. It is evident that, as for children with other cancer diagnoses, leukemia patients' quality of life is affected, by changed relationships with family members and friends and by decreased number of participation in social activities[39,40]. Furthermore, it is shown that leukemia patients must cope with a higher burden of disease, measured by patient-reported functioning than patients with CNS/solid tumors [41]. Besides, our findings of more and overall longer inpatient stays for deceased patients with leukemia is particularly relevant, since burden of treatment for children with leukemia might be underestimated given the comparably good prognosis for this diagnostic group[41] and leukemia being the most common type of childhood cancer. Such an underestimation might result from the so-called halo-effect, that is, the tendency to let one

key characteristic of a person or a situation (e.g. very good prognosis) outshine other characteristics (e.g. burden due to the disease and treatment)[42].

Regarding the percentage of time spent in the hospital during the entire duration of illness (inpatient-proportion) and the number of days being inpatient during the last month of life our findings reveal that our sample of leukemia patients spent more time in hospital than the other diagnostic groups, both during their entire illness and during the last month of their life. These results correspond to a study which reported longer last hospital stays for leukemia patients [23]. One study found an association between more than 10 hospital days during the last month of life and lower quality of care ratings by physician[43]. Against the background of relationship between longer hospitalizations and quality of care, the highly interrupted daily life[3,4], difficulties to maintain social relationships and accompanying isolation[9,10,12], and affected familial relations[6,9] especially apply to leukemia patients. Spending more time in the hospital not only decreases the odds of benefitting from protective factors, it also magnifies children's major stressors, for example, those associated with treatment procedures, loss of control and the hospital environment[14]. In addition to the higher burden due to treatment found in our study, another revealed that hospitalized children were also burdened by more symptoms than outpatients[44].

Finally, compared to the overall sample leukemia patients were less likely to die at home. Similar results were reported in two US studies[22,23]. Low home death rates are not problematic per se as a literature review concluded that evidence is lacking for the notion that most families wish for death at home[25]. With respect to our results, it needs to be further examined why leukemia patients are less likely to die at home and relevant other factors that determine place of death, such as sex of the child or educational level of the mother[24]. If parents of deceased leukemia patients or the children themselves are less likely to prefer home as the place of death, for instance, because of a higher stress and strain of providing

end-of-life care at home as compared to children with other cancer diagnoses, the lower proportion of death taking place at home can be attributed to parental or children's wishes and are therefore justified. On the contrary, if they show the same preference for home as the place of death as other parents and children, the treating staff should facilitate the realization of these preferences. In the latter cases, realization of parental (or children's) preferences is vital because parental adaptation after a child's death is better when a child died at home and parents whose child died in the hospital reported higher ratings of depression[45,46]. However, lower home death rates for leukemia patients may have resulted from the fact that this diagnostic group was more likely to die because of therapy-related complications, but not due to progression of the disease which hinders the timely transition to palliative care at home [23,47]. In fact, it was shown that in cases of progressive cancer higher efforts were made to suggest home as the place of care and, if wanted, of death[23]. Lastly, in our sample 53% of the children who received palliative care died at home. This home death rate is comparable to home death rates reported in other studies from Brazil with 59%, Australia with 61%, and England with around 40%[24,48,49].

Although burden of treatment is a currently emerging field, it is still lacking specific research as well as theoretical development[27,28]. Our findings reflect antecedents of burden of treatment, namely treatment characteristics of children who died due to cancer. Being confronted with a higher burden of treatment, pediatric leukemia patients and their families are particularly vulnerable. For example, more hospitalizations mean more interruption of daily life, longer hospitalizations lead to a higher likelihood of changes in relational networks and more absence from work or school. These effects, in turn, negatively affect both antecedents (e.g. support by a social network) and consequences of burden of treatment (e.g. quality of life, adherence). From this perspective, it can be argued that a high burden of treatment is associated with less favorable health care outcomes[26].

Burden of treatment theory argues that patients' ability to participate in the provided treatment, termed "agency", is dependent on social networks that support individual agents, termed "relationality"[29]. Furthermore, the authors identify a group of persons, not the individual patient, as the adequate unit of analysis[29]. When thinking about burden of treatment of pediatric cancer patients, both notions are particularly relevant because pediatric patients are highly dependent agents. They have to rely on their parents for mobility, finances, and participation in treatment-related decision-making. Additionally, the child's cancer disease becomes a life-changing experience for the whole family, calling for family to be the unit of analysis. A higher burden of treatment for children who died of leukemia translates itself into a higher burden for the entire family unit. Therefore, health care professionals could aid in reducing the burden of treatment when they are better aware of the characteristics of leukemia treatment and by constantly applying this awareness to the entire family.

Limitations

This study has several limitations. First, the analysis of medical records is necessarily limited to both the available information in the records and to the quality of the information. Second, it is possible that the extracting research assistants may have overlooked relevant information. Both these factors may have contributed to missing values which, in turn, resulted in different sample sizes for some of the analyses. Researchers tried nevertheless to gather all relevant information by thoroughly extracting, and by asking the collaborating physicians for support. Third, since data collection took place at different centers, information in the records is not presented in a consistent manner which could have affected the extracted data. Fourth, medical records were written in French, German, and Italian and extracted in English. Accordingly, inter-extractor agreement with respect to linguistic

nuances could be limited. However, researchers discussed a set of extractions and continued discussing throughout time of data collection. Fifth, our sample of deceased children is subject to a selection bias. However, we think that our results on number and duration of inpatient stays can be transferred to some extent to the sample of children with cancer who would survive.

Finally, retrospectively examining the duration and number of inpatient stays naturally does not capture the quality of these hospitalizations that could be indicated by the work of the psychosocial team, by the access to spiritual care or by the palliative approach within a hospital. Besides, exclusively focusing on the location of end-of-life care and of death does not obtain a complete picture of the quality of end-of-life care which requires considering additional factors such as the palliative concept of the respective ward, the parental preferences, demographic characteristics or collaboration between inpatient and outpatient settings. Generally, more time spent in the hospital does not necessarily indicate lower quality of life for the child. On the contrary, in some cases hospitalizations may be experienced positively. For example, an inpatient setting can evoke a feeling of certainty on the part of the family, allows psychosocial and spiritual care, facilitates certain activities and therapies that are not available at home, or provides relief for caregiving parents. Capturing these indicators of quality of care and quality of life cannot be achieved by a retrospective analysis of medical records, but necessitates a prospective qualitative study which addresses the experiences of the patient and the family. Nonetheless, as shown above current research seems to validate the view that inpatient stays represent burdensome experiences for the child and the family. Besides, it is self-evident that higher numbers of inpatient stays eo ipso lead to more disruptive effects on families' daily lives with all consequences for social relationships, leisure activities, or psychological and physical well-being discussed above.

Therefore, interpreting number and duration of inpatient stays as indicators (not sole determinants) of burden due to treatment is legitimate.

Conclusion

Our findings reveal that deceased children with cancer and their families face a high burden of treatment. However, on average deceased pediatric leukemia patients and their families are burdened by more and longer inpatient stays, by a higher proportion of inpatient-stays, by spending more days in the hospital during the last month of the child's life, and by a higher likelihood of the hospital as the location of death. Their comparably good prognosis may be causing an unintentional underestimation of their burden of treatment. Health care professionals (in both inpatient and outpatient settings) who are involved in the treatment of this especially vulnerable patient group should, if possible, mitigate burden of treatment and shape treatment regimes in close collaboration with the family aiming for a minimum of disruption and of changed daily life. That is, the treating staff not only should keep treatment throughout the course of the illness (including end-of-life) on an effective and necessary minimum, but also has to work towards understanding what kind of impact treatment and treatment-related aspects have on family's and children's daily lives[26]. Indicating burden of treatment, our findings enable physicians and nurses to better grasp and understand what is imposed upon the patient and the family, for example, in terms of inpatient stays and accompanying factors. Finally, burden of treatment was analyzed in this paper using objective variables extracted from medical records highlight the disruptive effects of treatment that are likely to have a great impact on their daily life and that go beyond exclusively focusing on side effects on the part of the child as indicators of treatment burden. Thus, it emphasizes the claim to think about burden of treatment from a family perspective.

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Chapter 5

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The Conceptual Understanding of Pediatric Palliative Care: a Swiss Healthcare Perspective

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Abstract

Background: Health care providers' perception of pediatric palliative care might negatively influence timely implementation. The aim of the study was to examine understanding of and attitudes towards pediatric palliative care from the perspective of health care providers working in pediatric oncology in Switzerland to promote the timely implementation of pediatric palliative care.

Methods: Five mixed focus groups were conducted with 29 health care providers (oncologists, nurses, psychologists, and social workers) at five Swiss pediatric oncology group centers. The focus group interviews were analyzed using thematic coding.

Results: Most participants associated pediatric palliative care with non-curative treatment. They regularly reported difficulties in addressing palliative care services to families due to the strong stigma surrounding this term. They also thought that the notion of palliative care is very much linked to a policy context, and difficult to reconcile with children's everyday life. To overcome these obstacles many participants used synonyms such as comfort or supportive care. A few providers insisted on the need of using palliative care and reported the importance of positive "word of mouth".

Conclusions: The use of synonyms might be a pragmatic approach to overcome initial barriers to the implementation of palliative care in pediatrics. However, this tactic might ultimately prove to be ineffective as these terms might acquire the same negative connotations as palliative care. Positive word-of-mouth by satisfied families and healthcare providers might be a more sustainable way to advocate for pediatric palliative care than replacing it with a euphemistic term.

Background

Since its introduction in 1975 the term palliative care (PC) has been subject to fluctuations in meaning[1-5]. For almost two decades the term has been used interchangeably with hospice, end-of-life or terminal care[3]. In 1990 the World Health Organization (WHO) shifted away from the "end-of-life" mindset by stating that palliative care is applicable earlier in the course of illness, in conjunction with anticancer treatment. This definition was further amended in 2002 when PC was uncoupled from prognosis and the target population was broadened to include patients facing a life-threatening condition[2,5].

The current gold standard approach in PC, as defined by the World Health Organization (WHO), consists of the *concurrent* administration of curative treatment and PC with attention to patients' physical, psychological, social, and spiritual needs. The definition provided by the WHO[6] is said to be a philosophical, rather than a dictionary definition[7] as it does not just report what "palliative" literally means ("the alleviation of suffering"), but invokes certain values (e.g. patient-centeredness, holism and multidisciplinarity) that are aimed to guide action and improve practice[1,8]. The principal aim of PC is to maintain the quality and the meaningfulness of life for both patients and their families. Unlike hospice care, PC is not limited to terminal care, but is appropriate for patients in all disease stages[6].

Within pediatrics and in pediatric oncology the early implementation of PC has been widely endorsed[9,10] as it has been associated with improved survival and quality of life[11-17]. According to the "Cancer Pain Relief and Palliative Care in Children" document[18], pain management for children should start *at diagnosis* and continue throughout the course of illness alongside curative treatment. Despite these recommendations, patterns

of late referral continue to persist in pediatric oncology[16]. As a result, many children – like many adult patients – do not benefit from pediatric palliative care (PPC) or at least not in a timely manner[19]. Clinical reality appears thus to lag behind the paradigm shift made within PPC guidelines[19-21]. This suboptimal integration of PPC leads to a paradox: if the early introduction of PPC is beneficial to the child's and family's well-being, then why is there such reluctance to implement this principle in medical practice[22]?

Studies exploring barriers to PC have focused mainly on PC in adult healthcare[23-31]. Given that the group of pediatric patients with PC needs is considerably smaller compared to the adult patient group, fewer studies, with some noteworthy exceptions, have focused on barriers to PC in pediatrics[20,32-34] and evidence specific for pediatric oncology is rather limited[35]. Of these studies, the majority have used quantitative methods and/or focused on the perspective of a single type of pediatric healthcare provider. In all these studies family and patient reluctance to accept PPC is often cited as an important (perceived) barrier to early integration[13,32,36] and related to misconceptions of PPC goals due to its equation with death and dying[37].

Still, a recent survey study on pediatric cancer patients' and parents' attitudes toward early integration of PPC in oncology seems to debunk the myth that parents and children are not ready to integrate PPC[13]. Interestingly, the study found that for the majority of children and parents who participated in the study, the term "palliative care" was *unknown*. In the survey, the researchers described the PPC team as "a group of clinicians with expertise in symptom management and a goal of improving quality of life". These findings suggest that patients' and parents' attitudes toward PPC integration is influenced by the way in which PPC is explained[28] and that (in) adequate understanding of PPC by healthcare providers may bias families' attitudes and decisions towards the integration of PPC.

In Switzerland the knowledge gap regarding palliative care among lay people is still great[38] and awareness of children's unique PPC needs is scant compared to other countries like the UK[39]. Those who are familiar with the notion of palliative care associate it with a type of care that focuses on quality of life when faced with an incurable illness. A recent retrospective analysis of medical records of deceased pediatric patients has shown that late and non-referrals are still very common in the Swiss oncology setting and that a dualistic model of curative and palliative care continues to prevail[40]. Since health care providers' perception of PPC might influence that of parents and children, the present study aimed to examine the understanding of and attitudes towards PPC in Switzerland from the perspectives of health care providers working in pediatric oncology centers.

In order to gain a richer, more detailed account of staff members' attitudes toward PPC and to identify possible socio-cultural factors that influence their perceptions, we conducted focus group discussions with various stakeholders in the field of pediatric oncology: oncologists, nurses, psychologists, and social workers. The study goal is particularly important given that health care providers might *unwittingly* associate PPC with end-of-life care even if they claim to support the early introduction of PPC[32]. Thanks to their interactive nature, focus groups provide access to data that might be less easily obtained through surveys or individual interviews as some thoughts can only be probed within a group context[41]. Furthermore, in order to develop appropriate interventions that im- prove PPC provision within the field of pediatric oncology, it is important to listen to different care providers, especially in the Swiss context where PPC is usually provided by the primary oncology team rather than by PPC specialists. Since divergence of opinions on PPC is not uncommon among team members and might lead to an inconsistent message about PPC in interaction with families[42], we

intentionally used a *mixed* focus group approach. Finally, it is increasingly recognized that qualitative insights play an important role in closing the policy-implementation gap[43-45], this is particularly relevant for the PPC context where (conceptual) implementation barriers continue to persist despite the development of educational programs, PC's increased focus on quality of *life* (rather than on death) and its overall beneficial outcomes.

Methods

This study is part of a larger project on end-of-life decision-making in pediatric oncology where (a) surveys were carried out with physicians and parents and b) interviews were conducted with children suffering from childhood cancer, their parents, and physicians; and (c) a retrospective data collection from medical records was performed[40,46-48]. For this qualitative part of the study, mixed focus groups were conducted with health care providers at five of the nine Swiss pediatric oncology group centers (SPOG): three out of six centers in the German-speaking part of Switzerland, and both of the two centers in the French-speaking part. The remaining four SPOG-centers decided not to participate in the focus groups due to lack of time. Approvals were obtained from the respective cantonal research ethics committees.[1]

For each SPOG a reference person was identified who helped with participant recruitment and focus group scheduling. The reference person informed all members about the overall aim of the focus groups and their confidential nature. Upon consent of the participants, the recruiter then gave the research team a list of team members who had expressed interest to participate. Each of these persons received the participant information sheet from a member of the research team via email, immediately after recruitment and then again (as a reminder) some days before the actual date of the focus group discussion to give participants enough time to read over the participant sheet. The research team sent a request to all participants to find a common time to carry out the focus group discussions. Since initial recruitment happened with the mediation of a reference person it was difficult to establish how many persons refused to participate (mainly because of lack of time).

Focus groups were carried out by the first and second author between August 2016 and

June 2017 at the SPOG centers at a time agreed upon by all participants. At the time of the interviews, the first author was a postdoctoral researcher with a background in philosophy and ethics. The second author was a doctoral student with a back- ground in psychology and ethics. Both researchers had experience with conducting interviews. The total number of participants for each discussion varied between 4 and 8 healthcare providers. Oral informed consent was sought from all participants prior to the start of the focus group and registered upon consent. From an ethical point of view, for minimal risk research involving interviews studies with health care professionals whose data (transcripts or questionnaires) are anonymized, oral consent and active participation are ethically considered sufficient and proportionate. Furthermore, in Switzerland interviews with health care professionals (not patients) are outside of the human research act and do not require ethics commit- tee approval. To make sure that our experts were clearly informed, at the beginning of the discussion, the moderator briefly restated the purpose of the overall project, their role in the project and allowed participants to ask questions.

A semi-structured interview guide framed each focus group discussion. The guide was built on the data obtained from a retrospective review of medical records of children who died at the SPOGs between 2008 and 2014[40] and on the experiences of the research team during prior phases of the overall project. In order to fine-tune the questions the interview guide was evaluated by a collaborator working at a SPOG. Questions included information about (a) the participants' personal understanding of PPC, (b) the institutional attitude towards PPC, (c) discussions and communication processes with parents and children regarding PPC, (d) perceived obstacles to the implementation of PPC and (e) institutional referral practices. Questions on topics (c) to (e) were discussed with reference to a specific recent case that the team encountered. Most of the data presented in this paper derives from the questions related to topics (a), (b) and

(d) as they deal with *conceptual* barriers. The other topics will be analyzed in a future manuscript.

The five focus groups lasted between 90 and 120 min. One team member was the moderator; another team member was the co-moderator who took notes, kept track of time, and helped in asking follow up questions. To facilitate qualitative analysis, all the discussions were tape-recorded and transcribed verbatim in the language of the interview (German or French). Transcripts were returned to participants for revision. The 5 focus group transcripts were checked for accuracy by three researchers and transferred into the qualitative analysis software MaxQDA (version 12) to support the analysis process[49]. Three authors independently analyzed the transcribed data by reading the interviews several times. After a close line-by-line analysis of the transcripts, provisional categories were identified by each of the three researchers. In a next step – to ensure consistency in the analysis of the data – the three team members discussed their respective categories and reached an agreement about the coding scheme and superordinate themes across the different focus groups were developed. In a final step, the first author reviewed the group level thematic taxonomy through the eyes of each individual participant to see which themes represented and did not represent the individual's account. For this purpose, everything a single participant said was, first, marked in a specific colour and then re-read and compared to the themes discussed by the group as a whole. In this way, both differences and commonalities among participants were identified together with the overall context that triggered their claims. This step was important given that the focus groups were mixed and we wanted to do justice to the concerns of each individual and provider type. Saturation of data was reached after the 5 focus groups.

Results

A total of 29 persons (working at 5 different SPOG centers) participated in this study: 14 nurses (among which 2 nurses specialized in the field of PPC), 10 physicians, 4 psycho-oncologists/psychiatrists, and 1 social worker (see Table 1).

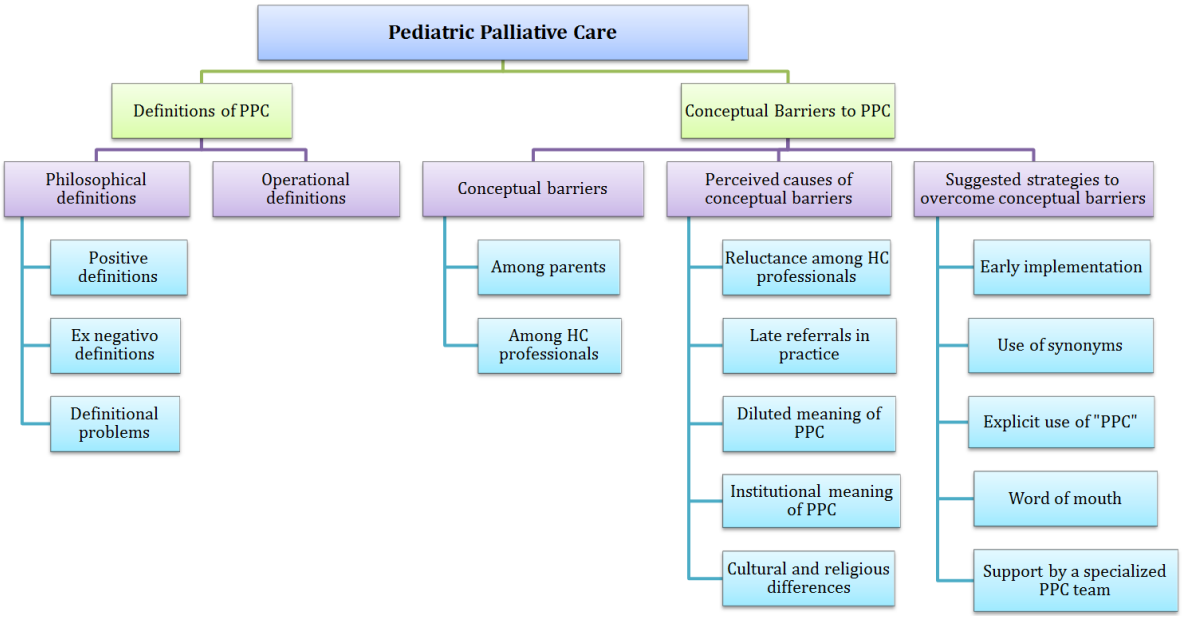
Table 1. Participants characteristics (N = 29)

All participants	N (Percentage) / Mean (SD)	
Gender (women)	21 (72.4%)	
Experience in years	14.9 (8.9)	
Professions	Experience in years Means (SD)	Gender in percent (women)
Nurses (n = 14)	16.3 (9.8)	92.9%
Physicians (n = 10)	16.1 (8.4)	30%
Psycho-oncologists / Psychiatrist (n = 4)	7.5 (4.1)	100%
Social worker (n = 1)	12	100%

Analysis with regard to the conceptual understanding of PPC by pediatric oncology care providers identified 2 major themes (and several subthemes): (1) Definitions of PPC and (2) Conceptual barriers, their causes & possible solutions. To improve clarity, the themes and their respective subthemes are presented in Fig. 1.

To illustrate the reported results, representative quotes were taken from the various interviews using pseudonyms for the participants as well as the center. All quotes were translated from German and French into English, and translations were checked for accuracy by two authors. Quotes will be presented in tables so that all three perspectives on a particular topic are easier to compare and also to provide an insight into the richness of experiences and opinions gathered.

Figure1. Themes



Definitions of PPC

Philosophical definitions of PPC

There was overall consensus on the common principles and values of PPC. In fact, most participants listed features that are essential to the gold standard of PPC: a family centred and active approach to care, offered by a multidisciplinary team and incorporating physical, psychological, social and spiritual needs to ensure the best possible quality of life throughout the illness trajectory (see Table 2.1, section a).

In addition to these definitions that describe what PPC is or should be, many participants also provided "ex-negativo" definitions, that is, they used expressions to describe what PPC is *not* (see Table 2.1, section b). Most providers insisted on the fact that PPC is not the same as end-of-life care or care provided when death is imminent. However, at the same time the majority of the participants insisted on the fact that *within the Swiss oncology* context, it is best to provide PC when there is no response to curative treatment. Among them many believed that understanding palliative care as non-curative care was crucial in order for the team "to know where

they stand" and to be able to communicate with the parents in a clear and honest way (see Table 2.1., section b, ii). Despite this shift, various participants insisted that this is not an abrupt one, but a gradual process which allows everyone involved to accept the fact that the child is dying. There was one participant who explicitly disagreed with the description that PC starts when there is no hope for cure. Within this regard it is important to highlight that participants were aware of the existence of other, often contradictory, definitions of PC (see Table 2.1, section c). Although most participants agreed that the provision of PPC is closely connected to the absence of cure, this rather broad understanding of PPC raised important challenges when implemented in practice.

Operational definitions and operationalization of PPC

Many participants acknowledged that clear guidance was missing on an operational level (see Table 2.2) and that this led to divergence of opinions among team members, especially with regard to the question of *when* to introduce palliative care. A physician reported (speaker 5, center 1): «There is always, or very very often, a moment when – I do not know how many times I have nurses heard saying: "but why do they [physicians] continue?"». Nurses recounted that they sometimes have the feeling that physicians push the boundaries too far and implement PPC too late. A nurse (speaker 4, center 1) reported how upset she was when she forbade a young patient to eat the sausage of which he was so fond, while knowing that he might only live for another 3 months: «We are the ones who enforce the [food] prohibition (…) that's maybe why we start asking ourselves questions more quickly».

On the other hand, nurses acknowledged that they do not always have the same medical information as the doctors, and that this can create divergence. They thus noted that it is important to sit together so that everybody is on the same page. Some

participants expressed the concern that these disagreements might be due to confusion about the meaning of PPC: «Sometimes there is no agreement because the definitions are unclear» (nurse, speaker 4, center 5).

Table 2. Definitions of pediatric palliative care

1. Philosophical definitions	
a) Positive definitions	<u>i.) PPC is gold standard (WHO)</u> For me palliative care means comfort care, holistic care, it means taking care of the child and his/her family in a physical and psychological sense; total care of the entire family**.** (*Nurse, speaker 2, center 1*) It really means «not to abandon» … to provide palliative care is something very active, to stand by all the life projects until the very last second. (*Nurse, speaker 1 with PPC specialization, center 2*) The most important thing is really the quality of life and (...) to create a network for families, for the whole family (...) and important is also to collaborate and also to have a multidisciplinary team. (*Nurse, speaker 3, center 3*) For me it is important that the persons who cared for the patient before do not just disappear and leave the patient alone (…) but that the contact remains until the end and even beyond the end, ideally. (*Physician, speaker 1, center 5*)
b) **Ex-negativo definitions**	<u>i.) PPC is not end-of-life care</u> To me it is quite clear that palliative care is something for the child (…) within a comfort approach and with the aim of increasing the child's quality of life. But it does not necessarily imply the imminent end-of-life. (*Nurse, speaker 4, center 2*) End-of-life care that starts normally during the last four weeks, but PPC starts of course much earlier. (*Physician, speaker 1, center 4*) ii) <u>PPC is not curative treatment</u> [PPC] is the total care of a pediatric patient from the moment one knows that the oncological disease is incurable. (*Physician, speaker 5, center 1*) In some other areas of PC it is different – in the case of a chronic condition the child cannot be cured anyway. In oncology, however, we always start with a curative treatment, but then there are situations (…) in which we say: we cannot cure the child (…) and then there is a switch to palliative therapy, which can take a long time, but then we know that the child will die of either of the illness or the treatment. (*Physician, speaker 1, center 3*) The shift from curative to palliative, I think that in Switzerland that is very different from the UK where palliative care teams are integrated from the time of diagnosis (…) until there is still hope for a cure, that they will find a treatment. Here [in Switzerland] they will not consider them [patients] palliative. (*Nurse, speaker 2 with PPC specialization, center 2*). <u>iii.) PPC is not an abrupt shift</u>

	It's true that from a medical point of view there is never an abrupt shift from a curative to a palliative approach, there is always a transition phase (…) often the family and the patient, but maybe physicians too need such a transition time before coming to terms with the fact that the child will not heal. It's not something on/off. (*Physician, speaker 5, center 1*) The decision-making regarding the shift from curative to palliative treatment, is a process (…) in which one goes back and forth, in which one is not fully certain … it is not a fixed point in time. (*Psycho-oncologist, speaker 5, center 4*)
c) Definitional problems	i.) Disagreement on the meaning of PPC I do not agree. For me curative and palliative are not in opposition, or consecutive, they are often rather concurrent, especially in the case of our patient group [pediatric oncology] where once the one aspect then the other can be more important. (*Physician, speaker 3, center 4*) ii.) Acknowledgement of various definitions I believe that you can understand many different things under palliative care and that is precisely the problem. (*Physician, speaker 2, center 4*)
2. Operational definitions of PPC	
	i.) Uncertain timing due to definition The shift from curative to palliative is not that clear. From when can we say that a person is in palliative care? In practice this passage is not so clear to me. On the other hand what palliative care stands for I think is very clear here [to professionals in the hospital] (*Social assistant, speaker 2, center 2*) ii.) team discussions due to unclear definition I still find it really difficult to say that now a patient is palliative or not. (…) I really notice - that nurses and doctors often have a different view on the whole. Sometimes we do not immediately find a consensus. Maybe the concepts are not clear (…) I remember the last big roundtable we had over a difficult case in order to all have the same understanding and to bring in ideas in order to know: "what next"? (*Nurse, speaker 4, center 5*) It [PPC] was introduced quite late in the service (…) in some situations the children felt more and more uncomfortable, they had a poor quality of life and the nurses were the ones who shouted; "Help! " (…) (*Nurse, speaker 2, center 1*). The individuality within the team, the different values, the different attitudes towards life, are often not explicitly made (…) what remains unsaid can create barriers (…) it can lead to fractions or different fronts, without it being made explicit (*Psycho-oncologist, speaker 5, center 4*) iii) need to overcome conflicts Consensus is absolutely necessary (…) you have to try to find it. Which way do you want to go, how do you want to go there? (…) perhaps you need to get together two or three times. Even if you want to avoid conflicts (…) you have to face them (*Physician, speaker 1, center 5*).

Conceptual barriers, their causes and possible solutions

Conceptual barriers among parents and professionals

Many participants – across the different provider types – were convinced that parents are reluctant to start PPC be- cause they associate it with "death and dying", "loss of hope", and "giving up" (see Table 3.1, section a). According to a social assistant (speaker 2, center 2) the stigma sur- rounding PC is so substantial that «the term "palliative" is the biggest enemy» when addressing families. Some participants acknowledged that they also often "struggle" with the concept and have difficulty accepting the next phase in the child's illness course (see Table 3.1, section b).

The perceived causes of conceptual barriers

Some participants acknowledged that their own attitudes towards PPC might negatively influence those of families' and further compromise timely implementation of PPC (see Table 3.2, section a).

Another important barrier that was cited (mainly by nurses) for the wrongful association between PPC and death was that PPC is still implemented relatively late in the course of the illness, in response to a bad prognosis or when children's quality of life becomes very poor (see Table 3.2, section b). On the other hand, some participants seemed to be concerned that introducing PPC too early; at the time of diagnosis – as set out by the WHO guidelines – risks diluting the meaning of PPC (see Table 3.2, section c). If PPC is uncoupled from the actual dying process and the non-curative phase, then «we are all in a palliative situation» (physician, speaker 3, center 2) insofar we all will die 1 day and this will cause confusion. Some were concerned that by uncoupling PC from dying, one risks marginalizing an important component of PPC, namely bereavement care. «There is little notion (…) in oncology of bereavement care which is an essential part of palliative care» (Nurse, speaker 2, center 1). Others reported that all-round care is provided from the moment the patient enters the hospital, and wondered what difference introducing PPC earlier would really make (see Table 3.2, section c).

As well as issues related to the timing of PPC, participants also argued that the notion of PPC is very much linked to an institutional, political and professional context, but that this PC philosophy is difficult to reconcile with children's every day life: «In the child's personal context what does that mean, palliative care?» (physician, speaker 3, center 2) Various participants associated PC with adult, and in particular elderly, care and emphasized that also on the policy level PC was rarely addressed with regard to children (see Table 3.2, section d).

A final cause has to do with the cultural and religious background of patients and their families. When parents' background significantly differed from traditional Swiss culture, participants frequently reported difficulties with introducing and providing PPC. In many cases, it was the presence of pain which finally convinced the parents to implement PPC, but this was often at a rather late stage (see Table 3.2, section e).

Table 3. Perceived conceptual barriers and their causes

1. Perceived conceptual barriers	
a) **Among parents**	i.) PPC equals death, giving up, loss of hope There are parents where you have the feeling that they do not consent to the palliative care process because they think: «I am giving up on my child». (*Psycho-oncologist, speaker 4, center 3*) For many families ″palliative″ means ″death, whereas palliative care does not mean that you will die. (*Nurse, speaker 2, center 1*) The difficulty may be to address the issue of death, but it is also like (…) if we abandon all efforts. For parents this is hard, because they perceive it as "we give up" and it does not meet their expectations. (*Psycho-oncologist, speaker 7, center 2*) The last case was a patient who medically speaking was in very bad shape (…) the mother fought with all her strength against the slow change of [treatment] course [from curative to palliative]. She had the impression that we were denying the boy the chance to heal. (*Physician, speaker 7, center 5*)
b) **Among HC professionals**	i.) Struggle to accept a next phase There is much work to do regarding the wording; many things belong to us, to the caregivers, our difficulty to accept that we pass from a curative to a palliative phase. (*Psycho-oncologist, speaker 7, center 2*) The concept [of PPC]I s one thing, it is a bit the rational part, like safeguards

	and structures it offers support, orientation and security. On the other hand, you have the attitude of the department…how [team members] *actually* experience palliative care (…), what we live *(Psychologist, speaker 5, center 4)*
2. Perceived causes	
a) Reluctance among professionals	<u>i) HC professionals' fear of PPC</u> If there is no pain yet or something that leads to palliative care (…) I feel it as an obstacle myself. We are afraid of pronouncing the word and at the same time we do not know how to tell it differently. (*Physicians, speaker 5, center 2*)
a) Late referral practice	<u>i) PPC as the grim reaper</u> We only talk about PPC at the last minute; it's like we sign the child's death warrant (…). So maybe if we were introduced before, it [PPC] would not have the same effect on the family (*Nurse with PPC specialization, speaker 1, center 2*) If we take the definition of palliative care, and ask ourselves whether death is probable, unavoidable within "six months", so the speak (…) then I think we do not anticipate the situation enough and then all of a sudden it declines very quickly most of the time. *(Nurse, speaker 1, center 1)*
b) Diluted meaning of PPC due to integrative approach of WHO	<u>i) No clear distinction leads to confusion</u> It's very hard to have a discussion and to state "not everything is lost yet" and at the same time mention "palliative care". (*Physician, speaker 5, center 2*) <u>ii) Total care is always already provided</u> Holistic care is there already (…) that's why there is confusion, I think. But ok, that is the WHO definition. Comprehensive care is present throughout the illness course, whether it is in the palliative or curative phase. (*Nurse with PPC specialization, speaker 1, center 2*) The concept [PPC] disturbs me (…) I think we have been providing medical care in a humane way for centuries and I do not think (…) care has to be renamed (…) the patients must receive best supportive care from to beginning until the end of life. (*Physician, speaker 7, center 5*) What would be the added value, which is currently missing, if one would integrate it [PPC] from the diagnosis onward? (…) there is already a total care approach (…) I think there is not necessarily an advantage. (*Physician, speaker 3, center 2*)
c) Institutional meaning of PPC	<u>i.) The term PPC is not related to the life-world of children</u> I think the word ″palliative″ (…) is inadequate in pediatrics because if we place a person of 50 or 60 years old in palliative care, we know that the person will die at an age close to the natural age of death with a palliative care treatment of 10-20 years depending on the type of illness. In the case of a child (…) we will not be reach the natural limit. (*Nurse, speaker 4, center 1*) We knew that talking about palliative care might constitute a barrier to certain families, but for the public health sector who finances us, it was a must (…) we needed to call ourselves [the service] ″palliative″. (*Nurse with PPC specialization, speaker 1, center 2*) An important topic is the difference between the life-world of adults and the life-world of children (…) when one in Switzerland talks about palliative care or about its political or infrastructural aspects then one has to be aware that the child is not taken into consideration. (*Physician, speaker 2, center 4*)
d) Religious & cultural	Something that makes it difficult to address palliative care is that 80% of the immigrant population comes from far away (…) and have many different

differences	cultural backgrounds (…) What does palliative care mean for a Swiss person, but also in the broader sense in the world. Culturally speaking, what does it [PPC] mean? (*Nurse, speaker 4, center 1*)

Suggested strategies to overcome the conceptual barriers

The participants provided several strategies to help overcome the conceptual barriers. Some of them suggested introducing PPC earlier so that this type of care would be part of the "landscape" and not be considered to be the "grim reaper" (Nurse, speaker 1 with PPC specialization, center 2). Timely implementation of PPC was believed to promote a more natural transition and take away the a priori negative connotation of PPC (see Table 4.1, section a). Several healthcare providers suggested rebranding PPC by using a synonym which is more easily understood and less stigmatized, such as comfort care, best supportive care or "*accompagnement*"[2] as these words do not directly relate to death and dying. They believed that these more commonly used terms better expressed the caring goal of PPC (see Table 4.1, section b).

A few participants, however, insisted on the importance of using "palliative care" because they feared that the use of a euphemism would not really help overcoming the stigma associated with the word. They further emphasized that it was healthcare providers' task to pro- mote the values of PPC among policymakers and the general public. The use of the term "palliative care" was also said to be crucial to give a clear indication of the patient's overall treatment and care goals and thus to have a common language and understanding within the team (see Table 4.1, section c).

A few participants also reported about the importance of "word of mouth": families who are satisfied about PPC can help overcoming the taboo surrounding this type of care and become PPC advocates (see Table 4.1, section d).

Finally some care providers highlighted the importance of having an external specialized PPC team as this could offer an additional perspective on the situation and provide

support to the primary team, who might be too emotionally involved to start PPC in a timely manner. On the other hand, in some centres, concerns were raised about whether different teams could successfully work together (see Table 4.1, section e).

Table 4. Strategies to overcome conceptual barriers

1. Strategies to overcome conceptual barriers

a) Early implementation of PPC	i) Timely introduction Perhaps the fact of intervening sooner would create less fear. When people hear "palliative care" they immediately think: "Ah that means: death". (*Nurse, speaker 8, center 2*)
b) The use of synonyms	ii) Euphemisms I like the term "accompagnement" in these situations (…) it means "being with" (…) "palliative" evokes, in my opinion, something a priori negative and I do not feel comfortable using it. (*Nurse, speaker 3, center 1*) For me, more than using this word [PPC], it's more about contextualizing it together with the parents and the child. I think that the child and the parents have a different view on the situation. We must try to make them understand that knowing that a disease incurable does not mean stop giving care, but entails a different way of caregiving (…) one should use the word "comfort care", for me that is more appropriate (*Nurse with PPC specialization, center 2*)
c) Explicit use of the term PPC	i) De-bunk the stigma We need a common language (…) You have to call a cat ″a cat″ (…) among the general public there is maybe a kind of ″mystification″ of palliative care, in the sense of palliative equals death, but it is our task as professionals to finally inform the public on the federal level and the rest. (*Nurse, speaker 1, center 1*) ii) Bring clarification to family & team The concept (…) should bring clarification to the treatment team and the patient. Therefore I actually understand palliative care as non-curative care. That is a crucial point for me, which I then need to discuss with the parents so that we know where we stand. (*Physician, speaker 2, center 4*) We on the nursing level use the word (…). We say: «the doctors have talked with the parents, we are in a palliative care situation now». I think for us this word signifies a change of attitude in the perception of the type of care we will provide. (*Nurse, speaker 4, center 2*)
d) "Word of mouth"	i) Family satisfaction We deprive ourselves a little of the opportunity to give them a chance (…) In see the family room I see how much families speak to each other. Every time a family has really benefitted and been satisfied, well, this information goes around. Families speak a lot among them *(Social assistant, speaker 2, center 2)*
e) Support of specialized PPC team	i.) Benefits of specialist PPC team I think we would benefit to have a systematic support [external PPC service] (…) to bring a bit of order in the discussion, in the ideas, in the emotions when preparing to pass from curative to palliative care. (*Nurse, Speaker 3, center 1*)

	ii.) Drawbacks of specialist PPC team There are different models in Switzerland and in the world, also in pediatrics, on this subject matter, with some hospitals having specialized teams in the background (…) I think that it is not desirable (…) there are not so many cases in oncology (…) it would be an exaggeration to have such a team (…) it would also create certain problems. (*Physician, speaker 1, center 4*)

Discussion

Early PPC is encouraged by various international oncology organizations as it has been associated with improved symptom management and quality of life of both children and their families[21]. Still, to date, many patients who could benefit from PPC services do not receive them, or at least not in a timely manner[50]. Conceptual confusion has been identified as an important barrier to adequate implementation of PPC. Given the influential role of healthcare professionals on families' decisions, this study offers a unique insight into the personal understanding of and attitudes towards PPC among various types of pediatric oncology providers. There is only a small body of literature on barriers to PC in pediatrics and most of the existing studies have used quantitative research methods. However, in order to better understand the "paradox" in PPC – the persistence of late and non- referrals despite PC's beneficial impact – qualitative research might be more appropriate as it enables a more complex insight into people's opinions and motivations.

The study results show that most participants recognized the important value of PPC and had good knowledge of its core principles and objectives. The interesting finding is that although they clearly distinguished PPC from end-of-life or terminal care, many of them – across the various provider types – insisted that, within the pediatric oncology context, PPC is best provided when curative treatment is no longer an option. Thus, although the WHO guidelines embrace an integrative approach from the time of diagnosis, most participants defined the target group for PPC in a much more restrictive sense. However, they also emphasized that the transition from curative to PPC is not abrupt, but is actually a rather gradual process during which both families and healthcare providers slowly but steadily become

aware of the need to redirect the care provision. Understanding PPC as non-curative was considered necessary to generate this change in attitude. Some participants expressed the concern that "mixing" the two care approaches might cause confusion among both family and staff members and thus be counter-productive.

While most participants supported PPC principles, they were worried about the lack of guidance on implementing them within clinical practice. Timely integration of PPC continued to pose a problem on the operational level and led to conflicts among the team members. Due to their daily contact with patients and families at the bedside, participants believed that nurses are more pro-active than physicians in encouraging PPC. Various interviewees recognized their own personal difficulties with the transition of treatment goals and welcomed the intervention of a specialized PPC team to lessen the burden. However, not all participants were in favour of such a specialized PPC team. Some believed that the primary oncology team could offer all the necessary PPC and seemed to be concerned about possible interpersonal conflicts. Research has shown that such conflicts are not unusual as the non-hierarchical structure of PPC tends to challenge the traditional hierarchical culture of the medical system[51,52].

Confirming the results of previous studies[22,24,26,30,31,53], participants regularly reported difficulties in addressing PPC services to families due to the strong (perceived) stigma surrounding this word. To overcome this obstacle many participants adopted a euphemistic term, such as comfort care, supportive care or *accompagnement*[2]. This finding confronts us with the following paradox: given that the definition of PPC has evolved considerably over the last decades and its *ascribed* (formal) meaning in the guidelines is highly beneficent ("improvement of life quality"), why then is the *lay* meaning still overly negative[1,2,22]? Prior studies suggest that negative attitudes towards PPC among patients and families are often influenced by staff members' own negative

image of PPC[25,32,33,36]. Some of our participants seemed to confirm these findings and this might confirm the idea that like many Western societies, Swiss society is still death-denying. Still we believe that given the centrality of death and dying in the Swiss public debate, it is difficult to maintain that this topic is still taboo. Hence, it is crucial to better understand care providers' own perception of PPC to comprehend their aversion of the term palliative care. Our results show that various participants personally disliked the term "palliative care" not, or at least not primarily, because of its strong association with death or dying (many of them, in fact, were in favour of understanding PC as non-curative!) but be- cause they considered it to be a concept that is incompatible with children's everyday life. In this context, it is interesting to reflect further on the meaning of the terms, "support", "comfort" and "*accompagnement*". They are all words that are widely used in daily life to describe a willingness to be with others, to stand by someone, to be present[54]. This ordinariness stands in sharp contrast with the term "palliative care" which was perceived by the providers as being too policy-loaded. This finding is worth exploring more fully as it can provide novel insights into the conceptual barriers impeding implementation of PPC and thus be used to frame the design and analysis of future empirical studies on this topic. Although we cannot address this topic extensively here, we should keep in mind that both on an institutional level and in the public debate, PC is often discussed and promoted as a life- and choice-affirming alternative to euthanasia and physician assisted suicide as PC intends neither to hasten nor postpone death and patient preferences are heavily promoted in the WHO definition of PC[6,55]. This means that PC is often debated within a context of autonomy and choice in which planning towards and acceptance of death are actively encouraged. This may explain why some participants considered the concept of PC to be out of touch with children's perspective. By using synonyms such as com- fort, support and "*accompagnement*" they

might have wanted to reinforce PC's original aim of being close to patients and families in care and affection.

Some participants were skeptical about rebranding PPC. They insisted on endorsing the term and debunk- ing its negative meanings by introducing PPC earlier. In this way families will be less scared to revise the treatment goals the moment their child is not doing well. Other participants, however, critized this integrative approach. They argued that holistic care *is* provided at the time of diagnosis and wondered what the added value would be of introducing PPC at the start of the illness trajectory. In line with other studies, the question that we have to ask is: if PPC is no longer a special type of care, but becomes part of the medical "mainstream", independent of any advanced prognosis, then does it not become too de-coupled from death[2]? Some authors are concerned that in order to improve acceptance, PPC guidelines have become death-sensitive and risk marginalizing those for whom PC was developed in the first place: dying children and their bereaved families[56-59]. A few participants seemed to share this worry when they reflected on the limited bereavement care possibilities in pediatric oncology.

A small number of participants addressed the possibility of rebranding PPC by relying on positive word-of-mouth. Studies have shown that word-of-mouth communication might have a great influence on the healthcare behaviour of the general public, especially in our current era of internet-based communication and the in- creasing use of social media[60].

A last concern expressed by the participants was the interaction with patients and families from a different, non-Swiss cultural and religious background. At present, there is little or no research on how ethnic and religious minorities perceive and understand PC definitions[61].

Limitations

First, because of the mixed focus group approach and the possible power differences that go along with it, it is possible that not all participants freely expressed their views on the subject matter, so some issues might have been left unsaid. Furthermore, the familiarity among the participants (most of them were part of the same team) might have resulted in taken-for granted "party-line" attitudes regarding PPC[62]. However, we intentionally used such an approach to mirror the actual "natural" group dynamics in the Swiss SPOGs where PPC is usually provided by the primary oncology team, which includes nurses, psychologists, psycho-oncologists etc. in addition to physicians. This inter professionalism might have allowed the production of in- sights that could have been less accessible in single provider type groups. Moreover, the moderators encouraged all participants to engage actively during the debate. Finally, the advantage of using an acquaintance group was that participants could provide more details regarding certain events or experiences and challenge the statements of other participants if they did not correspond to the actual hospital practice[62]. Second, given the specific Swiss pediatric oncology context, findings are not generalizable to other contexts abroad with specialist PPC teams and a different healthcare system. Third, since the focus group data collection was part of a bigger project that has been running since 2012, a few of the study participants knew the re-search project and the team already. Thus, we cannot exclude that the responses of some participants might have been influenced by their perception of the overall goal of the project. Finally, because most of our participants were women, our findings might be gender biased. However, since the workforce in both the pediatric and palliative care context is predominantly female, our sample does reflect the setting that we wanted to examine.

Conclusion

Despite important changes in the formal definition of PPC, it has still a overly negative connotation in the minds of many parents and healthcare providers. To counter this trend, calls have been made to initiate PPC at diagnosis and if necessary, rebrand the term PPC as supportive or comfort care. Many participants in this study seemed critical about the "from diagnosis onward" directive and clearly associated PPC with non-curative treatment. To most of them, the adequate timing of PPC remained a major challenge. Although the philosophical definition of PPC leaves room for patient individuality, it complicates clinical practice as it does not provide clear protocols. More referral tools are needed to help oncologists to identify children and families with palliative care needs. Further, although PPC has increasingly profiled it- self as being concerned with the patient's quality of life (rather than with death) this shift has not overcome all stigmas. Therefore, perhaps the conceptual obstacle to PPC is not so much death itself, but the way in which PC is discussed on both a policy level and in public debates, that is, in terms of choice, autonomy and personal development. This interpretation could find support in the fact that our participants considered the term "palliative care" to be out of touch with the child's perspective and preferred to use synonyms that are closer to PC's original aims: to offer support to patients and families in pain, anger, sadness and laughter without any normative expectations. Other well-designed empirical studies are needed to further explore these findings. Still, although the use of these alternative terms might be a useful and pragmatic approach to overcoming the *initial* stigma[30], in the end, it might be ineffective as these words might gradually acquire the same negative connotations as PC as long as there is no change in the public discourse on dying. The best way to counter this trend may be to promote (on- and offline) positive word-of-mouth[63] among satisfied families and health care

providers. By sharing their stories, families and healthcare providers could become the true ambassadors of PPC. More research is needed on how healthcare professionals can use online word-of-mouth on social media for PPC advocacy. Also, further efforts should be pursued to develop PPC educational and training programs for healthcare staff (including conscious self-reflection). Finally, critical reflection is needed on the possible practical and conceptual shortcomings in PPC guidelines themselves, in order to better support PPC healthcare providers.

Endnotes

[1] Switzerland is a federal state which is divided into 26 cantons. Each canton has its own cantonal constitution, approved by the federal parliament. For the current study, we obtained the approval of 4 research ethics committees.

[2] It is difficult to adequately translate the French word “accompagnement” in English since it is deeply indebted into the Francophone culture. The notion emphasizes patients’ embeddedness in society, rather than putting the focus on their individuality. The best translation would maybe be “accompanying”, “to be a companion”.

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Chapter 6
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Palliative Care Initiation in Pediatric Oncology Patients: A Systematic Review

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Abstract

Palliative care (PC) aims to improve quality of life for patients and their families. The World Health Organization and American Academy of Pediatrics recommend that PC starts at diagnosis for children with cancer. This systematic review describes studies that reported PC timing in the pediatric oncology population. The following databases were searched: PubMed, Web of Science, CINAHL, and PsycInfo databases. Studies that reported time of PC initiation were independently screened and reviewed by 2 researchers. Studies describing pilot initiatives, published prior to 1998, not written in English, or providing no empirical time information on PC were excluded. Extracted data included sample characteristics and timing of PC discussion and initiation. Of 1120 identified citations, 16 articles met the inclusion criteria and comprised the study cohort. Overall, 54.5% of pediatric oncology patients received any palliative service prior to death. Data revealed PC discussion does not occur until late in the illness trajectory, and PC does not begin until close to time of death. Despite efforts to spur earlier initiation, many pediatric oncology patients do not receive any palliative care service, and those who do, predominantly receive it near the time of death. Delays occur both at first PC discussion and at PC initiation. Efforts for early PC integration must recognize the complex determinants of PC utilization across the illness timeline.

Introduction

The World Health Organization (WHO) released a seminal report, titled *Cancer Pain Relief and Palliative Care in Children*, in which it recommended that palliative care (PC) for children with cancer ought to begin at diagnosis, irrespective of prognosis[1]. Other international health organizations – the American Academy of Pediatrics (AAP)[2], Institute of Medicine (IOM)[3], European Association for Palliative Care (EAPC)[4], and the Royal College of Paediatrics and Child Health (RCPCH)[5] – have all since adopted a similar recommendation[6]. These calls for earlier PC are grounded in evidence of an unmet need: the high illness burden and degree of suffering are well established among children with cancer[7]. Additionally, numerous studies have demonstrated that the tight prognostic limits of hospice – a type of PC reserved for the end-of-life – are incompatible with the full spectrum of physical, psychological, social, and spiritual needs[8-10]. PC presents an effective solution as both children with cancer and their parents report significantly enhanced quality of life from PC involvement[11,12].

With the improvements in medical therapy for pediatric oncology patients[13], children now survive for longer periods and require extended PC, making pediatric PC an increasingly important area of research[14,15]. Recent studies have investigated related ethical issues: how and when children should be involved in decision-making[16-19] and what disparities exist in PC access[20,21]. Moreover, various studies have demonstrated PC for children with cancer is initiated late in the illness trajectory[22-24], indicating a discrepancy between the normative recommendation for early integration and referral practices in pediatric oncology. Yet, no systematic review of the timing of PC initiation has been conducted to compile this growing body of literature.

Understanding the current state of PC timing is necessary to inform efforts to expand PC access, increase the time that children benefit from PC, and better support pediatric oncologists. As such, the purpose of this study was to systematically review literature describing the current timing practices of PC initiation in children with cancer. The two key events involved in the start of PC are the initial discussion with or without specialist consultation and the first instance of palliative service provided. If PC services started close to time of death, it is important to know if discussion occurred early and PC was deemed unnecessary, or if discussion also occurred late. Knowing the specific timing of these events will identify where in the care continuum barriers may lie. Thus, this review sought to answer a guiding questions: a) what time elapses between cancer diagnosis and PC discussion or consult; b) how long before death does PC discussion occur; c) what is the PC duration received before death, and d) what proportion of children receive PC.

Methods

Search methodology

This systematic review of literature on the timing of PC in pediatric oncology patients was performed in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analysis (PRISMA) guidelines[25]. Details of the systematic review protocol were registered on the PROSPERO International Prospective Register of Systematic Reviews (CRD42018108557). We searched PubMed, Web of Science, CINAHL, and PsycInfo for publications between 1 January 1998 and 15 December 2017, examined the citations of included articles for relevant additions, and solicited additional citations through discussion with topic experts. The year 1998 was selected as the beginning date because the WHO declared in this year that PC for children with cancer ought to begin at the time of diagnosis[1]. The WHO declaration altered the paradigm of PC initiation, so practices prior to this date may not be comparable. We developed the following Boolean search phrase based on controlled vocabulary results from the Medical Subject Headings (MeSH) terms database: (Palliative OR Hospice OR End-of-Life) AND (Pediatric* OR Child* OR Adolescent* OR Teen*) AND (Cancer OR Oncology OR Tumor* OR Neoplas*) AND (Duration OR Start* OR Time death OR Timing death OR Begin OR Began OR Time referr* OR Timing referr*). The terms “hospice” and “end-oflife” care were included in our search because researchers and healthcare staff commonly equate them with PC, but care was taken during screening and full-text review to ensure the reported data reflected the first iteration of PC. If the patients in the included study had not previously received PC, then that study was included because the described hospice or EOL care also represented the first time PC was provided. Our search yielded a total of 1220 titles and abstracts across all 4 databases (Table 1).

Table 1. Search terms and search results on timing of pediatric palliative care

No.	Search terms	Matches PubMed	CINAHL	PsycINFO	Web of Science
1	Palliative OR Hospice OR End of Life	121.923	41.833	28.926	136.362
2	Pediatric* OR Child* OR Adolescent* OR Teen*	2.104.073	414.163	631.834	1.529.829
3	Cancer OR Oncology OR Tumor* OR Neoplas*	2.585.401	268.727	76.791	2.552.665
4	Duration OR Start* OR Time death OR Timing death OR Begin OR Began OR Time referr* OR Timing referr*	822.311	103.473	185.668	1.810.823
5	1 And 2 And 3 And 4	704	55	81	380

Note. Date of last search: 12th of August 2018

Exclusion criteria

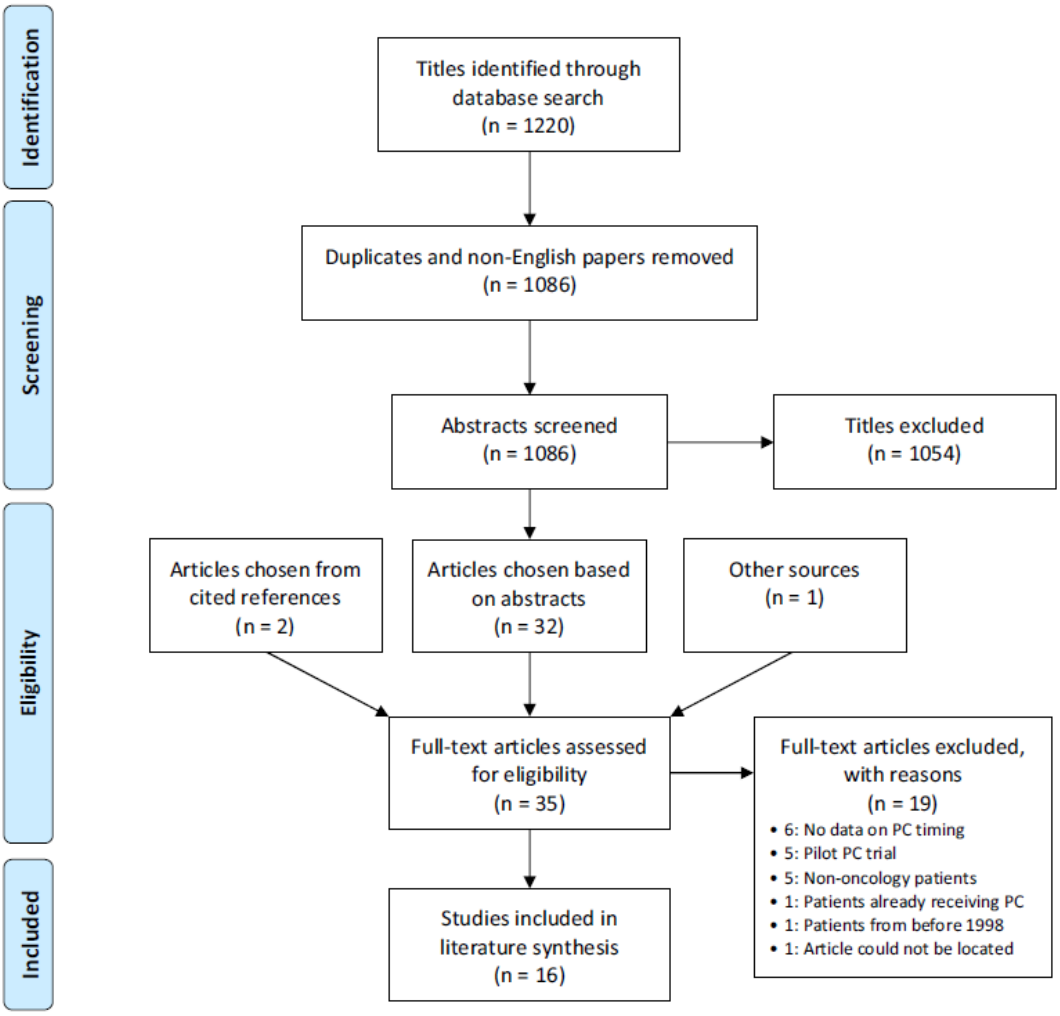
We defined the following a priori exclusion criteria: a) results published after the 1998 starting date of our search, but based on data collected from patients prior to the 1998 WHO recommendation; b) neonates because the type, presentation, and management of neonate cancer differs from those of older children[19], c) case studies because these describe exceptional medical situations and would skew our review; d) pilot initiatives that report results of a focused trial to encourage earlier PC consultation without pretrial data; e) no empirical data relevant to at least one of the primary study questions; and f) articles written in a language other than English.

Search results and data extraction

After duplicates were eliminated, two reviewers independently screened the article titles and abstracts to identify relevant articles. Of the 1137 unique citations identified, 1086 were published in English; titles and abstracts were independently screened by two authors, and 31 were selected for full-text review, in addition to three articles identified from reference lists and expert consultation; 16 remained after full-text review and comprised the full study cohort (Figure 1). The authors independently collected the necessary data using a purpose -

built Microsoft Excel extraction form: publication information, sample characteristics, and timing information from each of the articles. The two sets of extracted data were compared to validate the accuracy, and disagreements were resolved by discussion and input of a third investigator.

Figure 1. PRISMA flowchart for inclusion of studies



Analysis

All evaluated outcomes were included in meta-analysis, including three timeframes – time from diagnosis to first PC discussion, first discussion to death, duration of PC provided and

proportion who received PC. A study was included in meta - analysis if it reported mean with standard deviation or median with arithmetic range or interquartile range (IQR). For studies that only reported median and range, mean and standard deviation were calculated based on algorithms presented by Hozo et al[26]. All time data were converted into days. Data integration was conducted using Hedges-Olkin weighting models for inverse variance[27]. Random effect models were fitted by applying restricted maximum likelihood estimation, and Forest plots were constructed to visualize the results. All statistical procedures were executed with the metafor package in R - statistical software[28]. In cases of significant homogeneity and sufficient number of studies, Lipsey - Wilson moderator analyses were performed to examine study (sample country [US/non - US], publication year, terminology [PC - only/non - PC], and sample size) and clinical characteristics (principle diagnostic group [solid tumor/blood cancer]) as potential reasons for variability[29]. A two - sided P - value ≤0.05 was considered significant for all analyses.

Results

Characteristics of studies describing PC initiation

Of the 1137 identified studies, 16 studies were included in our review. Mean sample size was 237.2 (standard deviation = 294.5) patients and ranged from 17 to 1208 patients. Included data describes 3796 pediatric cancer patients, including 1438 solid tumor and 1231 hematologic cancer patients. Fifteen articles performed a retrospective medical review of records to study the timing of PC initiation, and one study was a prospective observation across six institutions (Table 2)[30]. Metrics in eight studies were representative of the pediatric oncology population, while the remaining examined subsets of this target population. Timing was a primary outcome in all 16 studies. While all studies used the phrase "palliative care," it is important to note that six studies also used end - of - life (k = 4) or hospice (k = 2) care interchangeably with palliative care.

The 16 included studies were published between 2002 and 2018. The median publication year was 2014, indicating that half of the studies were published in the last 4 years. Most studies (k = 9) reported timing of PC in North America, four in Europe, two in Asia, and one in Australia. However, there was little diversity in World Bank income classification: 15 included from high - income countries and one from an upper-middle - income country.

Table 2. Study characteristics of publications included in review (n = 16)

Year	Authors	Country of data collection	Study method	Sample size	Represents Population	Terms used for palliative care	Study period
2002	De Graves et al[31]	Australia	Retrospective review	17	Yes	Palliative care	1999-1999
2005	Bradshaw et al[32]	USA	Retrospective review	145	No	End of life	2000-2001
2008	Menon et al[33]	Malaysia	Retrospective review	247	No	Palliative care	2001-2007
2011	Tzuh-Tang et al[34]	Taiwan	Retrospective review	1208	No	Hospice	2001-2006
2011	Feudtner et al[30]	USA & Canada	Prospective data collection	102[a]	Yes	Palliative care	2008-2008
2012	Johnston et al[23]	Canada	Retrospective review	273	Yes	Palliative care	2006-2009
2013	Jalmsell et al[22]	Sweden	Retrospective review	95	Yes	End of life	2007-2009
2013	Thienprayoonet al[35]	USA	Retrospective review	114	No	Hospice	2006-2010
2014	Vallero et al[36]	Italy	Retrospective review	39	No	Palliative therapy	2005-2011
2015	Levine et al[37]	USA	Retrospective review	277	No	End of life	2001-2005
2015	Vern-Gross et al[38]	USA	Retrospective review	134	Yes	Palliative care	2001-2005
2016	Levine et al[39]	USA	Retrospective Review	615	No	Palliative care	2007-2014
2016	Ullrich et al[40]	USA	Retrospective review	147	No	Palliative care	2004-2012
2017	Ananth et al[41]	USA	Retrospective review	125	Yes	Palliative care	2010-2014
2017	Hoell et al[42]	Germany	Retrospective Review	65	No	End of life	2009-2016
2018	Rost et al[43]	Switzer-land	Retrospective review	193	Yes	Palliative care	2008-2014

Representation of the target population is defined as whether the metrics reported by the study estimate the pediatric oncology population.
[a]Number of cancer patients isolated from n = 515 cohort.

Time from diagnosis to PC discussion

Time from diagnosis to first PC discussion was reported in three studies, which included 485 pediatric oncology patients (Figure 2). In our random effects model, the weighted mean time to PC consult was 509.6 (standard error (SE) [95% confidence interval (CI)]: 37.6 [435.9-583.4]) days. PC discussion did not occur at diagnosis as recommended by WHO and AAP

guidelines ($P < 0.0001$). Test for heterogeneity suggested large variation in effect sizes within these studies (Q[2] = 11.7, $P = 0.003$), and Higgins $I2$ statistic demonstrated 78.9% of variability is not attributable to sampling error. There was an insufficient number of studies for moderator analysis.

Time from PC discussion to death

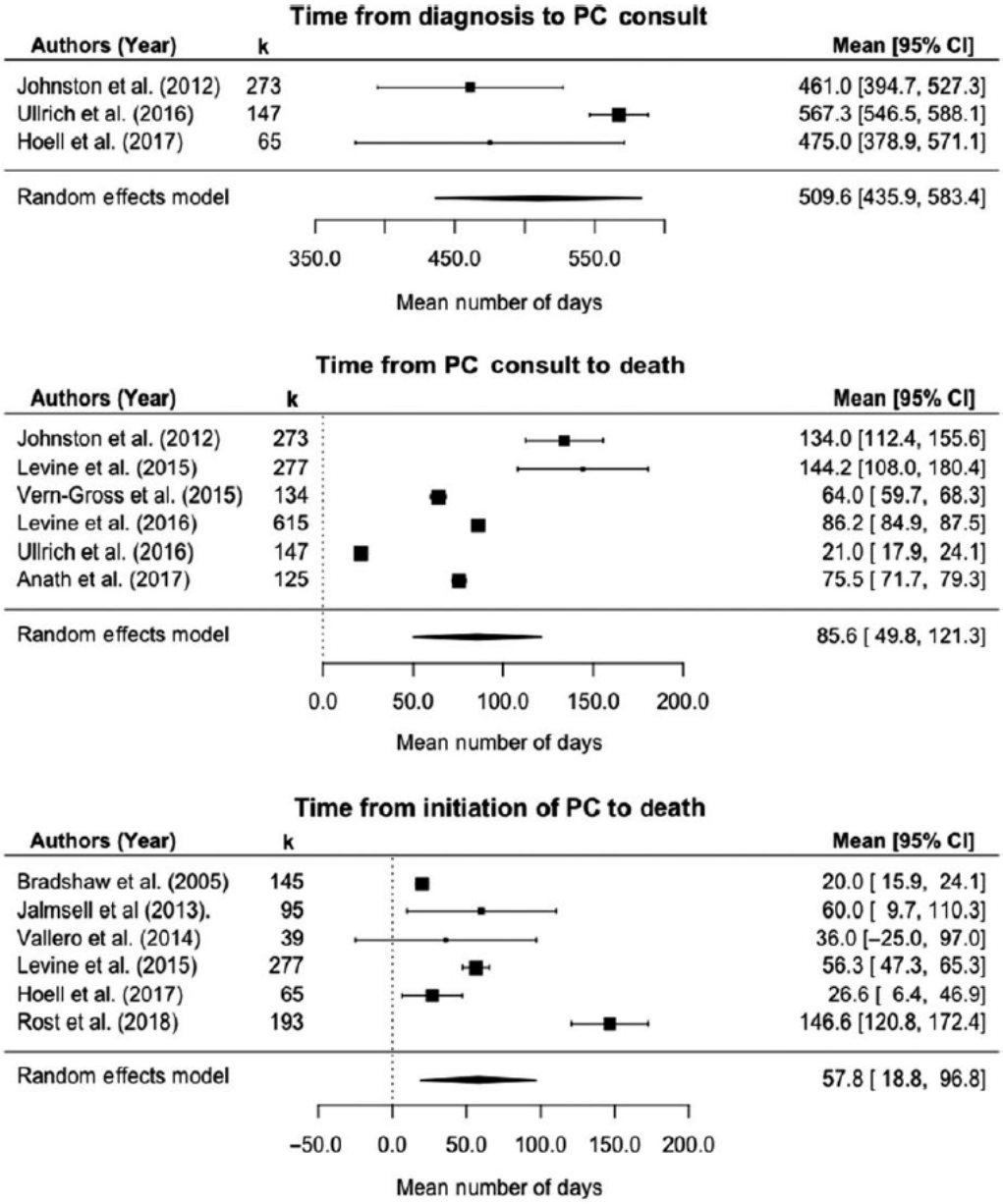
Six studies, including 1571 patients, reported time from PC discussion to death with a weighted mean length of 85.6 (SE [95% CI]: 18.3 [49.8 - 121.3]) days. Of note, this number is significantly smaller than the mean time from diagnosis to PC discussion, indicating that PC is discussed late in the illness timeline. An additional study reported median time was less than 37 days, but this study could not be included in our descriptive analysis because it did not report a measure of variation.30 In the two studies that reported both time from diagnosis to PC discussion and PC discussion to death, time from PC discussion to death was comparatively short, comprising 25.4% and 3.6% of the total illness duration[23,40]. Heterogeneity was significant for this outcome (Q[5] = 1547.6, P < 0.0001), and Higgins I2 statistic was 99.8%, suggesting much of the variability across studies was due to the heterogeneity. However, the role of individual moderators in this variation could not be investigated due to the small number of studies.

Duration of PC

Time from formal PC initiation to death was reported in six studies, including 814 patients. One additional study reported a mean PC duration of 69.4 days, but did not report a measure of variation and could not be integrated in our duration model[31]. In the random effects model for PC duration, mean duration was 57.8 (SE [95% CI]: 19.9 [18.80 - 96.8]) days. Two studies quantified both time from PC discussion to death and PC duration: Levine et al reported 144.2 and 56.3 days, respectively, while Vern - Gross and colleagues stated 64 and

31 days[37,38]. Taken together, these findings reveal initial PC discussion does not often result in prompt PC initiation. Effect sizes varied across studies (Q[5] = 134.8, P < 0.0001), and 97.8% of variability came from a source other than sampling error. Again, moderator analyses could not be conducted due to the small number of studies.

Figure 2. Timing and duration of palliative care provided to children with cancer

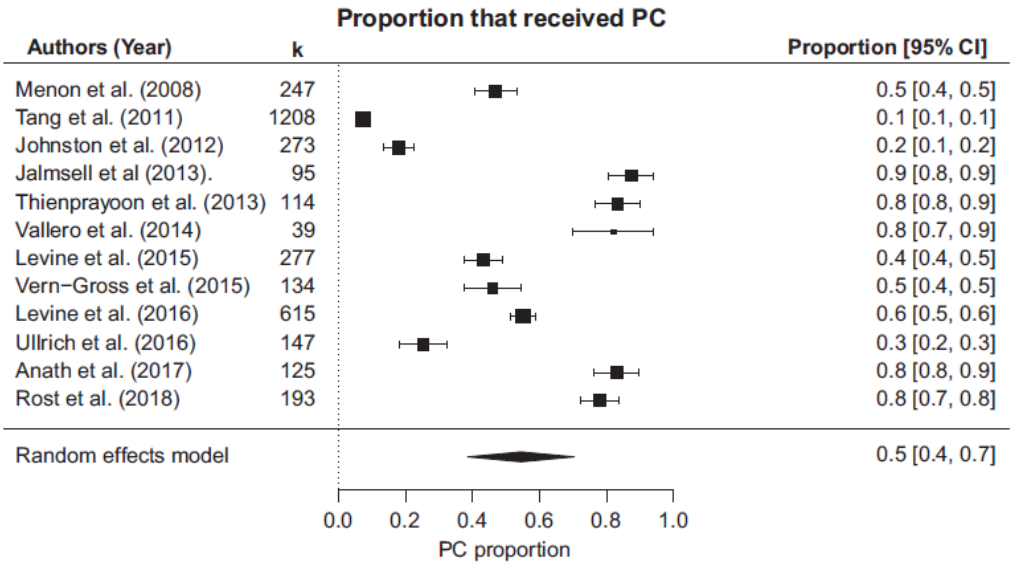


Proportion with PC

The weighted mean percentage of patients who received PC prior to death was 54.5% (SE [95% CI]: 8.2% [38.5% - 70.5%]) across 12 studies and 3467 patients. Higgins I2 statistic was 99.2%; test of heterogeneity was significant (Q[11] = 2230, P < 0.0001), and moderator analysis was conducted to investigate the effects of methodological and sample characteristics.

In mixed - effects models for moderator variables, increasing sample size was associated with a decline in proportion receiving PC (Q[1] = 6.1, P = 0.01). Mean PC proportions in US vs non - US studies were 56.0 vs 53.0 (Q[1] = 0.03, P = 0.86); PC proportion has not changed significantly over time (Q[1] = 1.4, P = 0.24). Studies exclusively using the PC terminology reported a mean provision fraction of 54.2%, compared to 55.1% in studies that also used EOL or hospice terminology (Q[1] = 0.003, P = 0.96). In subgroup analysis by malignancy type, weighted proportion was 39.9% (95% CI: 12.0% - 67.9%) in studies with predominantly hematological malignancies, 54.7% (29.7% - 79.6%) in those with mostly solid tumor patients, and 66.5% (27.1% - 100.0%) for studies in which principle cancer type could not be identified. Despite the variation, cancer type was not significant on moderator analysis (Q[2] = 1.3, P = 0.53) (Figure 3).

Figure 3. Proportion receiving palliative care in children with cancer



Discussion

This systematic review summarizes the timing of PC services for pediatric oncology patients reported in 16 publications published between 2002 and 2018. No systematic review has been conducted on the timing of PC initiation for children with cancer, and the present study is the first to compile the growing literature on pediatric oncology PC practices. Comparing PC provision within studies and across publications, it is apparent that PC integration was delayed at two time points: first PC discussion occurs late in the illness trajectory, and there is a delay between the initial conversation and start of PC. The growing number of publications and pilot initiatives demonstrate genuine energy to embed PC into conventional healthcare and improve the timing of PC discussion[37,44-46]. However, the results of this study suggest that PC integration at diagnosis remains an unmet objective.

Our finding that only 54.5% of pediatric oncology patients received any PC before death may suggest there are structural barriers that inhibit availability of PC. A multinational review of Children's Oncology Group institutions, which serve more than 90% of pediatric oncology patients in the US, found only 60% of providers offer PC services[47]. Potential reasons for the absence of PC services include lack of coordination between oncology providers and palliative programs outside the hospital, restrictive reimbursement models, and ambiguous roles of members in the care team[48]. While previous studies established the deficiencies in hospital infrastructure and our results demonstrate the effects on timing of PC provision, the drivers that cause hospitals to offer pediatric PC are not well understood. Further research is recommended to identify socioeconomic and geographic disparities associated with lower pediatric PC provision. Such research would identify areas where PC is underutilized in children with cancer and guide interventions to increase PC provision.

Another proposed explanation in literature for the lack of timely PC provision is the shortage of clinicians capable to deliver PC to children with cancer[49]. In a survey of PC providers – physicians, nurses, and other staff members – one-third of respondents cited insufficient training in PC as a barrier to earlier PC integration[50]. The limited exposure to targeted PC education in residency and fellowship means hospitals do not have staff with the training to meet the palliative needs of pediatric oncology patients[51-53]. Improvements have been made to increase training opportunities as the number of pediatric PC fellowships in the US have doubled since 2013[54]. However, adequate workforce availability remains a priority to expand the number of institutions that can offer PC to children with cancer.

Training in PC principles is not important solely for designated PC staff; communication by the primary oncologist has a critical role in initiating the first PC discussion with the patient and family. Physicians recognize the child's poor prognosis, on average, almost twice as early as parents do and thus, are often given the task to communicate the bad news to parents, explain treatment complications, maintain hope, and calibrate parental expectations[55]. Physicians, however, do not consistently articulate this prognosis effectively to the family of children with cancer: parents are more likely than their treating oncologist to indicate the primary goal is cure[56], and 61% of parents were more optimistic about their child's odds of a cure than the physician was[15]. Physicians view broaching PC as a stressor and delay the conversation, focusing instead on treatment arrangements[57,58].

Formal training is associated with feeling comfortable to manage end-of-life issues, but 75% of pediatric oncologists have not had any formal end-of-life training[59]. This communication deficit may cause parents to be overly optimistic and encourage them to pursue aggressive treatment until the physician is certain of their child's imminent death[56]. Communication skills trainings, supporting resources, and a team-based approach have shown promise as a means to facilitate earlier advance care planning and PC referral[60,61].

Multiple studies have cited conceptual confusion between PC and hospice or EOL care, as well as the stigma of hopelessness associated with PC, as significant obstacles to early PC discussions[6,62-65]. The International Classification of Disease coding system considers hospice and end-of-life care as synonymous with palliative care, and US clinicians are instructed to bill these services identically.[66] There are even definitional inconsistencies between and among PC guidelines of what palliative care constitutes[67]. If parents equate PC with EOL support, then it is understandable they will oppose the suggestion of PC at diagnosis or soon after. Yet, the Patient Protection and Affordable Care Act Section 2302 "Concurrent Care for Children" recognizes the difference between PC and EOL care and guarantees coverage under Medicaid or the Children's Health Insurance Program for concurrent hospice and curative care for all children under age 21[68]. Adoption of a new phrase without the same connotations of death may support physicians in patient discussions. While the use of new terminology in pediatric oncology has not been tested, a survey found that compared to "supportive care," the phrase "palliative care" was associated with decreased hope and increased distress in adult oncology patients and their families[69]. Furthermore, in a survey of 646 Canadian physicians, pediatric oncologists reported they would refer patients earlier if PC was renamed "supportive care"[70]. Additional research is warranted to devise methods to encourage earlier PC in the pediatric setting.

In moderator analysis, larger studies reported lower rates of PC utilization, suggesting that small single-institution studies may overestimate PC provision. Future research on the topic should consider sample size and setting when designing studies. Literature in which hospice or EOL terminology was used did not vary in results when compared to studies that only used PC terminology; this indicates that despite the connotational significance of terminology in the clinical setting, researchers frequently use these phrases interchangeably. We recommend

that researchers use caution to avoid confusion in reporting results and communicating findings to clinicians.

Finally, US location, temporal trend, and cancer type were not moderators of PC utilization. However, there was notable difference in rates between blood cancer and solid tumor. Pediatric blood cancers tend to have higher survival rates than solid tumors[71], which raises concerns that hematological cancer patients may receive more aggressive curative therapy until close to time of death. The low number of studies may explain why cancer type was not a significant co-variate in our moderator analysis, but additional research is warranted to further investigate a possible difference by malignancy type.

Limitations

Our analyses coalesce data across multiple sites and countries, which increase our confidence that these findings reflect true practice. All included studies were retrospective medical records reviews or prospective collection, which enhances the comparability across studies and reduces the chance of measurement error. Additionally, 12 of our 16 studies had sample sizes with $k > 100$. As such, we could calculate robust estimations of true effects. There are also limitations to our study. Inherent to systematic reviews, all included data were already published and may be affected by publication bias. Second, studies primarily originated from research in well-developed regions of the world. Research is needed outside of developed countries to better understand the state of PC access and availability for children with cancer worldwide.

Sample size may partially explain the variation in PC timing observed in our results. Yet, there are other potential moderators that were not reported and could not be tested. Culturally determined understandings of when a condition is considered terminal (eg prognosis smaller than 10% vs 5%) may affect timing in different settings. Other demographic (eg income,

ethnicity), clinical (eg comorbid chronic conditions), and methodological (eg different definitions of when PC began) characteristics may also provide valuable context for the observed results and thus, warrant further study. Aside from these limitations, this study is the first systematic review on the timing of PC and sets a foundation for efforts to improve quality of life for children with cancer.

Conclusion

Our results underscore that PC starts too late for children with cancer and are not in line with the recommended AAP, WHO, IOM, EAPC, and RCPCH guidelines[1,2,4,5]. Palliative care discussion does not occur until far into the illness, and PC does not start until much later. Each case is unique and must be evaluated using the caring physician's best medical judgment and with respect to the patient and family, but holistically, there is much room for improvement regarding PC timing for this patient population. Effective, timely communication maintains patient quality of life and dovetails the transition to palliative care, while poor communication may lead to poor treatment planning and psychological harms for the child and family[72]. Reasons for delayed discussion and initiation include insufficient resources and infrastructure, lack of training, and negative connotations attached to PC.

Findings in the present study regarding timings of PC discussion and initiation suggest pronounced obstacles across the PC lifecycle. As such, initiatives focused on specific referral points likely will not succeed. Programs designed to target PC timing must be robust and coordinated across the PC lifecycle to achieve effective improvement. Palliative care is central in pediatric oncology, and continued advocacy is

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Chapter 7

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Aligning Guidelines and Medical Practice: Literature Review on Pediatric Palliative Care Guidelines

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Abstract

Objective: Palliative care for children is becoming an important subspecialty of healthcare. Although concurrent administration of curative and palliative care is recommended, timely referral to pediatric palliative care (PPC) services remains problematic. This literature review aims to identify barriers and recommendations for proper implementation of palliative care for children through the looking glass of PPC guidelines.

Method: To identify studies on PPC guidelines, five databases were searched systematically between 1960 and 2015: Scopus, PubMed, PsycINFO, the Web of Science, and CINAHL. No restrictions were placed on the type of methodology employed in the studies.

Results: Concerning barriers, most of the papers focused on gaps within medical practice and the lack of evidence-based research. Common recommendations therefore included: training and education of healthcare staff, formation of a multidisciplinary PPC team, research on the benefits of PPC, and raising awareness about PPC. A small number of publications reported on the absence of clear guidance in PPC documents regarding bereavement care, as well as on the difficulties and challenges involved in multidisciplinary care teams.

Significance of results: Our results indicate that a critical assessment of both the research guidelines and medical practice is required in order to promote timely implementation of PPC for pediatric patients.

Introduction

Medical improvements have led to longer survival of pediatric patients[1,2]. As a result, the number of children with life-limiting and life-threatening illnesses is increasing. This explains why pediatric palliative care (PPC) is becoming an important subspecialty within the setting of overall healthcare. The illness of a child alters family relationships, and family members often experience psychological and emotional distress as a result[3]. Therefore, PPC aims to care for children and their loved ones in a multidisciplinary fashion. It focuses not solely on pain and symptom management, but also on the social, psychological, and spiritual well-being of patients and their families.

Palliative care developed first within adult cancer care. Its history is closely related to that of the hospice movement. For almost two decades, the terms "palliative care" and "hospice care" were used interchangeably, until the World Health Organization[4] fostered a conceptual distinction between the two[5]. Unlike hospice care, palliative care was no longer identified with "end-of-life care," but was "applicable earlier on in the course of illness, in conjunction with anticancer treatment"[4¨]. In 2002, the WHO promulgated a further amendment of the general definition of palliative care[6]. From then on, palliative care would be defined as an appropriate approach to care for *anyone* with a life-threatening condition *irrespective* of prognosis[7]. Other organizations have since adopted this broader understanding in their palliative care recommendations: the Institute of Medicine (IOM)[8], the European Association for Palliative Care (EAPC)[9] (the IMPaCCT standards), and the Royal College of Paediatrics and Child Health (RCPCH)[10].

The field of palliative care for children and neonates developed in the 1980s under the stimulus of palliative care for adults[11]. Still, it was mainly from the 1990s onward, with the formation of the Association for Children with Life-Threatening or Terminal Conditions and Their Families (ACT) (currently called "Together for Short Lives") and ChiPPS (the

Children's Project on Palliative and Hospices Services) that palliative care for children became more diffuse. "A Guide to the Development of Children's Palliative Care Services"[12] was one of the first documents to define palliative care as a total approach to care for children and the family[13]. "Cancer Pain Relief and Palliative Care in Children"[14] represented another significant milestone for the acceptance of PPC. This latter document states that pain management for children should begin at diagnosis and continue throughout the course of illness alongside curative treatment. Early integration of palliative care is highly beneficial for children with chronic and life-threatening conditions, as they often have complicated illness trajectories that come with a high degree of prognostic uncertainty[15]. This integrative model was soon also embraced by the American Academy of Pediatrics (AAP)[11,16].

Following the palliative guidelines (ACT, AAP, WHO), the current gold standard definition of PPC includes: (1) concurrent administration of curative and palliative treatment from diagnosis onwards, (2) attention to the physical, psychological, social, and spiritual needs of patients and their families; (3) provision of services 24/7 at home, in the hospital, or in health community centers that should not stop with the child's death; and (4) a team composed of at least physicians, nurses, psychologists, social workers, and family members. The main aim of PPC is to enhance the child's quality of life rather than focusing on the quality of the dying process[17].

Although the number of palliative care facilities for children has grown, it still lags behind the number of those for adults[18,19]. However, even when they are available, the existing guidelines are not adequately implemented. Many children who could benefit from palliative care services in fact do not receive them, or at least not in a timely manner[20-22]. A significant difficulty in the United States has been that, until the introduction of the Patient Protection and Affordable Care Act in 2010, palliative care for pediatric patients was determined following hospice regulations for adults, meaning that disease-related treatments were not covered and

reimbursement was limited to the last six months of a patient's life[23]. However, low referral rates have been reported in Canada[22] and in Europe as well[24,25]. Since the number of children with life-threatening and life-limiting conditions is still on the rise, it is even more important to address the underlying reasons for these late or non-referrals.

Various studies have identified barriers to palliative care in general[26-28] and PPC in particular[29,30], but mostly from the perspective of the patient, healthcare provider, or family member. None of these studies have documented these barriers through the looking glass of the PPC guidelines themselves. Therefore, our literature review had the following aims: to identify (empirical and theoretical) studies that discuss PPC guidelines in order to: (1) explore the development and evolution of these guidelines; (2) assess the barriers to their proper implementation; and (3) identify and address possible gaps in the guidelines.

Methods

A systematic literature review was completed by searching the following online databases: Scopus, PubMed, PsycINFO, the Web of Science, and CI-NAHL (table 1). The following search terms were combined using Boolean logic: "Pediatric*," "child*," "adolescent*," "palliati*," "palliative care," "hospice care," "guidelines," and "recommendations." The inclusion criteria were: (1) published between 1960 and May of 2015 and (2) written in English or German. A 50-year publication window was chosen to capture earlier studies that most likely utilized a different conceptual framework than more recent works (in particular, since WHO[4], AAP[16], and WHO[6]) when discussing and assessing palliative pediatric guidelines. No restrictions were placed on type of methodology (quantitative, qualitative, mixed-methods, or theoretical). In addition, literature reviews, abstracts, comments, conference proceedings, dissertations, and books were excluded.

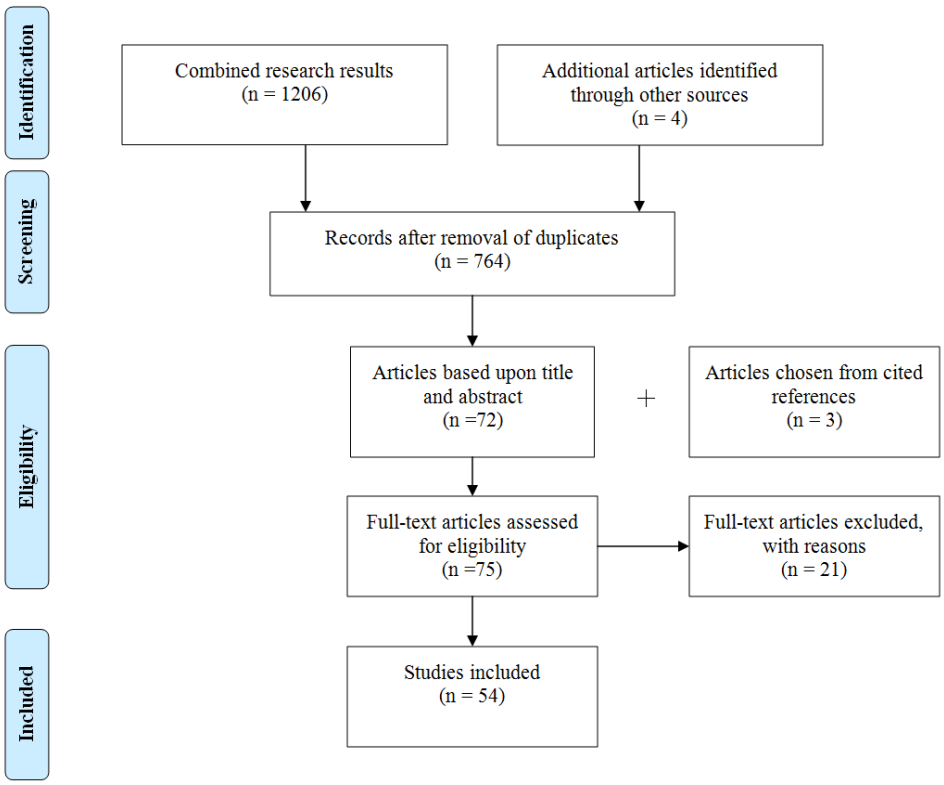
Table 1. Search terms

Nr	Search terms	Matches				
		Scopus	**PubMed**	**PsycINFO**	**Web of science**	**Cinahl**
1	Pediatric* or child* or adolescent*	2506694	3152041	215016	1057623	321967
2	Palliati* or palliative care or hospice care	37253	81110	20077	35105	25512
3	Guidelines or recommendations	471012	398677	131292	26327	93859
4	1 AND 2 AND 3	324	462	65	217	138

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) framework[31] was employed to organize the research (see figure 1), which resulted in 1,210 papers. After removal of duplicates, 764 remained. During the first step of the review, three

researchers screened all 764 titles and abstracts. All articles that *discussed* pediatric palliative guidelines (in general or a particular subaspect like pain and symptom management, psychosocial, spiritual, cultural, or bereavement care) were included. With respect to guidelines, we intended to deal not only with nationally and internationally recognized ones from pediatric organizations, but also those developed within a hospital context. The guidelines themselves (those issued by the AAP, WHO, etc.), however, were not part of the review, having been assessed in another publication (blinded for peer review). Studies that dealt exclusively with neonatal and/or perinatal palliative guidelines were excluded. Papers discussing one specific type of pain management (e.g., a particular drug or palliative sedation) or psychological support (e.g., music therapy) were also excluded. Discrepancies between reviews were evaluated by a fourth reviewer, who determined which articles were potentially eligible based on the abstract. In total, 692 articles were excluded.

Figure 1. Search process using a PRISMA systematic review of the literature



The reference lists of the remaining 72 papers were checked to identify any additional

studies. Three papers were added through this process. The final sample thus included 75 papers. During the next phase, the first author read the full-text versions of the articles. After evaluating each article, 21 were excluded because they (1) focused mainly on adult palliative care and only superficially on palliative care for children; (2) touched only implicitly on palliative care guidelines, addressing instead certain subaspects of PPC like decision making or the notion of a good death; or (3) focused in detail on certain pain medications.

To assess the remaining 54 articles, a data extraction framework was created with the following information: year of publication, country of study, methodology, references to PPC guidelines, type of PPC, barriers to implementation of guidelines, and recommendations for overcoming these barriers.

Results

Among the 54 papers included in our analysis, 29 were theoretical papers, that is, they proposed a theoretical framework of PPC or critically reviewed PPC models and practices. Of the remaining 25 studies, 10 employed quantitative methods, 10 qualitative methods, and 5 mixed methods. Most of the papers (*n* 46) were published after the 2002 WHO amendment. Some 30 papers were from the United States, 7 from the United Kingdom, 7 from Canada, and the remaining ones from Australia, Italy, Germany, Switzerland, Israel, and Poland, and one from two countries. The most frequently cited guidelines were WHO[14], AAP[16], IOM[8], and ACT (various editions between 1997 and 2013). Concerning the domains of PPC, 38 papers discussed palliative care in general. From the remaining 16 papers, 3 focused on two dimensions at the same time. The other 13 papers addressed one specific domain of PPC. End-of-life care was the main focus in a total of 21 papers, but only 4 of these dated from the most recent period, and 2 focused on bereavement care (table 2).

Table 2. List of included studies

Author, year, country	**Design**	**Participants**	**PPC Guidelines**	**Domain of PPC**	**Focus on EOL**
Ashby et al. (1991), Australia	Mixed method	Interviews: 15 HCP, 4 parents Questionnaires: 10 HCS, 2 parents	None	general	Yes
Baker et al. (2008) USA	Theoretical		IOM 2003, AAP 2000, WHO (1998)	general	No
Bergstraesser et al. (2013), Swiss	Qualitative	76 HCP	IOM 2003 and ACT 2009	general	No
Collins J. et al. (2002), Australia	Theoretical		WHO 1998	psycho-social and physical care (pain management)	No
Corr et al. (1985), USA	Theoretical		None	general	No
Dangel (2002), Poland	Quantitative	41 HCP	WHO 1998 and ACT 1997	general	No

Danvers (2003), UK	Mixed method	91 question. (parents + physicians); interviews (17 parents, 3 children) 7 focus groups (parents), 7 focus groups (Diana team), 86 multi-agency professional questionnaires	None	general	Yes
Davies et al. (2002), USA	Theoretical		Chipps 1998	spiritual care	No
Di Gallo et al. (2006), Swiss	Theoretical		None	bereavement care/spiritual care	Yes
Downing (2015), UK	Theoretical		WHO (2012: two step analgesic ladder) & ICPCN	physical care (pain management)	No
Eggly et al. (2011), USA	Qualitative	56 parents; 70 PICU physicians	AAP 2000	bereavement care	Yes
Epelman (2012), Brazil	Theoretical		Clinical practice guidelines for palliative care	general	Yes
Fernandez et al. (2011), Canada	Theoretical		WHO 1998	pain and psycho-social care	No
Feudtner et al. (2013), USA	Quantitative	126 HCP	AAP 2000	general	No
Foster et al. (2012), USA	Qualitative	1 case study	Clinical practice guidelines for palliative care	spiritual care	Yes
Frager G. (1997), Canada	Theoretical		None	general (but emphasis on physician care/pain management	No
Freyer et al. (2006), USA	Theoretical		None	general	Yes
Gowan (2003), USA	Theoretical		AAP 2000	physical care (pain management)	Yes
Heath (2009), Australia-USA	Qualitative	96 parents	IOM 2003	general	Yes
Houlahan (2006), USA	Quantitative	16 inpatient oncology nurses, 8 fellows	WHO 1998 - IOM 2002 - JCAHO 2001)	physical care (pain management)	Yes
Hubble et al. (2009), USA	Theoretical		AAP 2000	general	No
Janssen et al. (2004), Germany	Theoretical		ACT 1997	general	Yes
Jones et al. (2014), USA	Theoretical		AAP 2000 - WHO 1998	bereavement care	No
Kang et al. (2005), USA	Theoretical		AAP 2000/WHO 1996 (pain	general	No

			ladder)		
Kang et al. (2014), USA	Theoretical		AAP 2013	general	No
Kassam et al. (2013), Canada	Quantitative	75 parents ; 48 oncologists	Clinical practice guidelines for palliative care	general	No
Lee et al. (2008), USA	Qualitative	29 HCP	IOM 2003	bereavement care-moral distress staff	Yes
Liben (1996), Canada	Theoretical		None	general	Yes
Mandac et al. (2014), USA	Theoretical		AAP 2000, WHO 1998 and clinical practice guidelines	general	No
Matthews et al. (2006), UK	Qualitative	39 cases	ACT 2003 (RCPCH)	general	Yes
Maynard et al. (2014), UK	Quantitative	26 families, 60 hospice professionals and 53 HCP	ACT 2009 (RCPCH)	physical care (symptom management)	No
McNamara et al. (2008), UK	Theoretical		ACT 2005 (RCPCH)	general	No
Meyer et al. (2006), USA	Qualitative	56 parents	IOM 2003	general (focus on communication)	Yes
Mitchell et al. (2015), UK	Qualitative	8 physicians and 6 nurses PICU	ACT 2013	ACP	Yes
Moody (2011), USA	Theoretical		WHO 1996 - AAP 2000	general	No
Perilongo et al. (2001), Italy	Mixed method	13 cases	None	general	Yes
Postovsky (2004), Israel	Theoretical		AAP 2000	General (emphasis on DNR, palliative sedation, nutrition hydration, place of death)	Yes
Pritchard et al. (2011), Canada	Theoretical		WHO 1998	general	No
Remke et al. (2007), USA	Theoretical		WHO 1998	general (emphasis on socio-psychological and pain management)	No
Remke et al. (2012), USA	Theoretical		None	general	No
Rushton et al. (2006), USA	Mixed method	100 HCP	IOM 2003	general	No
Schmidt (2011), USA	Theoretical		AAP 2000 and IOM 2003	general	No
Shaw (2012), USA	Theoretical		WHO 2012	pain and symptom management	No
Sheetz et al. (2012), USA	Quantitative	65 parents	AAP 2000 - IOM 2003	general	No
Shelton et al. (2011), USA	Theoretical		AAP 2000	physical care (pain management)	No
Solomon et al.	Quantitative	211 physicians,	No	physical care	No

(2005), USA		116 house officers, 469 nurses			
Steele et al. (2008a), Canada	Qualitative	6 families, 4 HCP	ACT 2003	general (emphasis on respite care in hospice and on continuity of care)	No
Steele et al. (2008b), Canada	Mixed method	16 HCP	WHO - AAP 2000- IOM 2003	general (need research)	No
Stephenson (2000), USA	Theoretical		AAP 2000	general	Yes
Tan et al. (2006), USA	Quantitative	236 medical charts of deceased children	AAP 2000	general (emphasis on spiritual and pain management)	Yes
Thompson et al. (2009), USA	Quantitative	393 physicians	IOM 2003 - AAP 2000	general	No
Thompson et al. (2013), USA	Qualitative	2 case studies	WHO 1998 - IOM 2003	general (emphasis on pain management)	No
Toce et al. (2003), USA	Theoretical		AAP 2000	general	No
Twamley et. al (2014), UK	Quantitative	132 HCP	AAP 2000 ACT 2009 - IOM 2003	general	No

The Development of Pediatric Palliative Care Guidelines

To explore the development of PPC guidelines, we divided the studies into four different periods to correspond with the publication of the most important documents in PPC.

The 1980s and 1990s: The "Thirst" for Guidelines

We assessed four studies (one mixed-methods and three theoretical studies) that dated from *before* the 1998 WHO and 2000 AAP guidelines[32-35]. These papers welcomed the increasing trend toward honest and open communication about dying as a result of the hospice movement for adults, but stressed that the death of children was harder to discuss. They also underlined the importance of comfort and quality of life within the hospice philosophy and the need to include children in this type of care. They already introduced some of the central tenets of palliative care for children: the need for (1) total or holistic care; (2) bereavement care for

patients, family, siblings, and staff; (3) adequate pain assessment and control; (4) an interdisciplinary care team; and (5) continuity of care. Two studies focused exclusively on the needs of the *dying* child and did not make a clear conceptual distinction between hospice and palliative care[33,34]. Corr and Corr[32] defined hospice care as a form of palliative care for both terminal and chronic conditions, but failed to explain the difference. Frager[35] marked a kind of transition point, as this was the first paper among the four being discussed here to explicitly refer to an *inclusive* model of palliative care that should begin at diagnosis and be available not only for those with an imminently terminal condition, but also for individuals with a life-threatening disease, independent of outcome.

In the Direct Aftermath of Landmark Guidelines (1998 – 2002)

We assessed five papers[36-40] that were published shortly after or during the publication of internationally recognized palliative care guidelines[6,12,16,41-43]. Although most of them acknowledged the crucial role that guidelines play in promoting acceptance of the palliative care paradigm in pediatrics, some (empirical) studies raised the concern that these well-intentioned guidelines would remain purely theoretical in the face of the lack of national palliative care programs for children[40]. Stephenson[39] stated that hospice guidelines for adults (life expectancy of fewer than six months and no life-prolonging treatment) are too restrictive for pediatrics (due to difficulties involved with prognosis). Together with Collins[36] and Dangel[40], he embraced a total and integrative model ("from diagnosis onward") of palliative care, but failed to make a clear distinction between palliative and hospice care. Dangel[40] insisted that prognosis at diagnosis should be poor in order to initiate PPC. Perilongo and colleagues[38] equated palliative care with terminal care for the dying child.

The First Decade of Pediatric Palliative Guidelines (2003–2009)

The 23 papers dating from this period praised the increasing interest in palliative care within

pediatrics but underlined the limited availability of pediatric palliative services, as well as a lack of knowledge and specialized staff. Gowan[44] and Thompson and colleagues[45] highlighted the difference between palliative and hospice care. However, they noted that this adult model of end-of-life care is unrealistic for children with life-limiting and life-threatening conditions due to the restrictive policies regarding length and type of treatment (especially in the United States). In line with the guidelines, a growing number of articles underlined that, given the prognostic uncertainty, the pediatric population would benefit most from a holistic and integrative approach to care, that is, they insisted that palliative care should take place *alongside* curative treatment and begin at diagnosis[44-50]. This concurrent approach was also believed to improve the acceptance and thus implementation of palliative care guidelines for children[47,49,50]. At the same time, 12 of the 23 articles explicitly focused on PPC guidelines for end-of-life care[44,46,50-59]. Two of them[51,55] discussed guidelines for end-of-life homecare in which pain and symptom control, good coordination, and continuity of care, together with a partnership approach with respect to the family, are central tenets. Several papers emphasized the importance of honest and open communication with family members regarding prognosis and death[52,53,57,58,60]. Rushton and colleagues[61] addressed the problem of moral distress among healthcare staff due to competing professional ethical obligations and interdisciplinary conflicts. They proposed a facilitation model of education to support caregivers. Two papers offered guidelines for pain and symptom management[54,60]. Several (both empirical and theoretical) papers acknowledged the need to adapt guidelines to a child's individual needs, depending on age, maturity, culture, and religion[46,47,49,51,52]. The empirical study authored by Steele and colleagues[62,63] emphasized the need for more research on family experiences, pain management, and bereavement care in order to improve existing PPC guidelines. In the present sample, 12 of the 23 papers were empirical. Two qualitative studies by Heath et al.[53] and Meyer et al. [57] reported on parental satisfaction with end-of-life care and the need to

integrate their priorities (honest communication, emotional support, care coordination, integrity of parent– child relationships, and faith) to facilitate good PPC. Four other empirical studies focused instead on the experiences of pediatric healthcare personnel: two articles addressed the issue of moral distress[61,64]; two others concentrated on their preparation and training and concluded that, although staff members assessed themselves as knowledgeable; their awareness of the guidelines was limited[45,59]. The quantitative study of Thompson and colleagues[45] also reported that, although half of their 303 physicians (all AAP members) would refer patients before the end of life, very few of them would refer them at diagnosis. Their findings showed that implementing PPC in practice might be problematic. The authors expressed the need for a more practical service-related definition of palliative care to avoid any connotation of hospice care.

The Most Recent Period (2010–2015)

The 22 studies that dated from 2010 to 2015 reported that PPC is increasingly recognized as a priority by policymakers and hospital staff. Three theoretical studies conducted in the United States[7,65,66] referred to the 2010 change in federal legislation (the Affordable Care Act), which allowed for concurrent care (cure-related and palliative) for children until the age of 21. Still, despite recommendations and growing support worldwide, an important practice gap continued to exist between valued services and those that effectively reach the family[67-69]. In some of the world's most populous countries, PPC services are not readily available[70], and even within the same country there might be important differences in terms of number of staff, level of funding, and education, depending on the region or state[71]. Various studies expressed the need for further research on PPC in order to assess financial benefits[71]; improve pain management[70,72], show clinical benefits (improved quality of life and survival)[73], define familial needs, identify moral distress in members of the healthcare team, and explore the use and implications of advance directives in a pediatric population[7]. Almost all of these papers

highlighted the importance of providing holistic care that addresses the needs of all involved parties.

One qualitative study focused on spiritual care for patients and families[74]; five studies concentrated on bereavement care for the family during and after the child's death[7,73-76]; and five studies took into account the needs of healthcare professionals[67,70,73,76,77]. In line with the guidelines, various papers indicated that holistic care requires an interdisciplinary approach[70,73,74,76,78]. Another core concept supported by most of these papers is the idea that palliative care should be integrated into the routine care of patients with life-limiting and life-threatening conditions, from diagnosis onward, due to its beneficial impact on patients and families. Finally, two theoretical studies addressed the lack of and need for specific palliative care guidelines for adolescents[79,80].

Barriers to Adequate Implementation of PPC

Most of the 54 papers discussed the barriers to adequate PPC implementation, which can be subcategorized into eight different types of obstacles (table 3). The most common barriers discussed in these papers were connected to policy, operational, healthcare staff, and research factors. Other frequently occurring barriers to effective delivery of palliative care to children were associated with the uniqueness of the pediatric context: uncertainty about prognosis and lifespan make it difficult to determine which children should receive palliative care and when to start it. This uncertainty undermines communications between providers and families, who are generally more optimistic about a child's condition than the former and thus (perceived to be) hesitant about implementing palliative care in a timely fashion. Another insidious obstacle is the conceptual confusion between palliative and hospice care among both parents and healthcare providers. Interestingly enough, two recent papers[73,81] reported that, even in the case of adequate knowledge of and support for PPC principles, healthcare staff unwittingly

associate palliative care with end-of-life care, highlighting society's inexperience with childhood death. A limited number of manuscripts focused on conceptual and operational shortcomings within the guidelines (among others, see Bergstraesser et al.[73]; Jones et al.[76]).

Table 3. Barriers to pediatric palliative care

Barriers		**Related articles**
Policy factors – financial factors	Lack of human, structural, financial resources and medication	Bergstraessser et al. (2013); Collins et al. (2002); Dangel 2002; Downing (2015); Feudtner (2013); Kassam et al. (2013); Levine et al. (2013); Perilongo et. al (2001); Pritchard et al. (2011); Stephenson (2000); Thompson (2009); Toce et al. (2003)
	Lack of reimbursement	Baker et al. (2008); Gowan (2003); Mandac et al. (2014); Schmidt (2011); Stephenson (2000); Thompson (2009)
Operational factors	Lack of time	Baker et al. (2008); Davies (2002); Jones et al. 2014; Mitchell et al. (2015); Stephenson (2000); Toce et al. (2003)
	Lack of education – training - knowledge (communication skills, pain assessment and management)	Baker et al. (2008); Bergstraessser et al. (2013); Collins et al. (2002); Dangel (2002); Downing (2015); Eggly (2011); Gowan (2003); Houlahan (2006); Jones et al. 2014; Kassam et al. 2013; Levine et al. (2013); Liben (1996); Mandac et al. (2014); Mitchell et al. (2015); Moody (2011); Remke (2012); Rushton (2006); Schmidt (2011); Shaw (2012); Shelton et al. (2011); Solomon et al. (2005); Stephenson (2000); Thompson et al. (2013); Toce et al. (2003)
Healthcare staff	Attitude	Baker et al. (2008); Dangel 2002; Solomon et al. (2005); Thompson (2009); Thompson et al. (2013); Twamley et al. (2014)
	Moral distress – discomfort – grief	Bergstraessser et al. (2013); Eggly (2011); Jones et al. 2014; Lee et al. 2008; Liben (1996); Matthews et. al (2006); Pritchard et al. (2011); Rushton (2006); Thompson et al. (2009); Thompson et al. (2013)
Family factors	Fear of abandonment by oncology team	Epelman (2012); Pritchard et al. (2011); Thompson (2000); Thompson et al. (2013); Twamley et al. (2014)
	Cultural and religious values	Baker et al. (2008); Danvers (2003); Hubble et al. (2009); Mitchell et al. (2015)
	Misunderstanding of prognosis/treatment goals	Moody (2011); Thompson (2009); Thompson et al. (2013)
Societal factors	Death/dying taboo	Ashby (1991); Corr (1985); Liben (1996); Mitchell et al. (2015); Moody (2011); Stephenson (2000)
	Stigma/misunderstanding of PPC (hospice, give up etc.)	Kang et al. (2014); Pritchard et al. (2011); Stephenson (2000); Thompson (2009); Toce et al. (2003); Twamley et al. (2014)
	Treatment/cure oriented	Janssen et al. (2014); Kang et al. (2005); Remke (2012); Schmidt (2011); Stephenson (2000)
Clinical factors (Uniqueness of PPC)	Unpredictable disease trajectory and prognosis/Uncertainty when to start PPC	Bergstraessser et al. (2013); Freyer et al. (2006); Mitchell et al. (2015); Moody (2011); Pritchard et al. (2011); Schmidt (2011); Steele et al. (2008a/b); Thompson (2009)
	Limited number/scattered	Corr (1985); Danvers (2003); Mandac et al. (2014);

	population/rarity of death	Schmidt (2011); Stephenson (2000); Thompson (2009)
Research factors	Lack of evidence-based research	Feudtner et al. (2013); Foster (2012); Kang et al. (2014); Lee et al. (2008); Mandac et al. (2014); Moody (2011); Remke (2012); Schmidt (2011); Shelton et al. (2011); Steel et al. (2008b); Stephenson (2000); Thompson et al. (2009)
Guidelines factors	Lack of clear standards for some core aspects of PPC: PPC team; bereavement care; holistic care	Remke et al. (2012); Bergstraesser et al. (2013); Feudtner et al. (2013); Jones et al. (2014)
	Lack of clear referral criteria	Bergstraesser et al. (2013); Thompson (2009)

Recommendations to Overcome Barriers to Implementation of PPC

Seven areas of improvement were presented in the papers to help get around the barriers to PPC implementation noted above (table 4). Training and education of healthcare staff (and parents) were the most frequently listed recommendations to overcome operational barriers, although there were few references to precise teaching and training methods. Various papers identified training in pain and symptom management necessary to assess and alleviate pain in a timely manner and to confute the myth of opioid addiction among children. Education about the principles of PPC was considered crucial to avoid any conceptual confusion between hospice and palliative care. In particular, the principle of early integration (from diagnosis onward) was frequently cited as a way to make palliative care more acceptable. The underlying idea was that, if palliative and curative treatments are implemented at the same time, gradual transition of goals can occur. For the same reason, many studies emphasized the need to seek for the support of the PPC team (or specialized nurse) at an early stage. They could function as a kind of "glue" between families and primary care staff. Several papers also insisted on the importance of open and honest communication about prognosis and death and the need for guidelines to prepare parents and siblings for the child's death. Some articles spoke of the need for reflective practice among healthcare staff to address their (often unconscious) negative attitudes toward palliative care as end-of-life care. Four articles discussed advance care planning as a way to facilitate this process. Closely connected to this is the concern with

structured bereavement care (during and after a child's death) to support patients, families, and healthcare staff in their grief and moral distress. Two papers provided recommendations for spiritual care. Several insisted upon the need for evidence-based research in order to develop clear standards for various subfields of PPC.

Table 4. Recommendations for pediatric palliative care

Recommendations		**Related articles**
Team collaboration (PPC team, primary care team, parents and general practitioners)	PPC specialist support – integration in primary care team	Ashby (1991); Bergstraesser et al. (2013): Epelman (2012); Foster et al. (2012); Freyer et al. (2006); Hubble et al. (2009); Mandac et al. 2014; Maynard et al. 2014; Moody (2011); Postovsky (2004); Pritchard et al. (2011); Rushton (2006); Toce et al. (2003)
Training/education	PPC principles	Baker et al. (2008); Liben (1996); Dangel (2002); Danvers (2003); Downing (2015); Hubble et al. (2009); Kang et al. (2014); Kassam et al. (2013); Matthews et al. (2006); Mitchell et al. (2015); Schmidt (2011); Solomon et al. (2005); Steele et al. (2008a); Thompson et al. (2009); Toce et al. (2003)
	Pain and symptom management	Liben (1996); Frager (1997); Danvers (2003); Downing (2015); Houlahan (2006); Mandac et al. (2014); Janssen et al. (2004); Solomon etval. (2005); Thompson et al. (2013)
	Communication skills	Baker et al. (2008); Bergstraesser et al. (2013); Heath (2009); Hubble et al. (2009); Kang et al. (2005); Mandac et al. (2014); Meyer et al. (2006); Rushton (2006)
	Reflective practice	Downing (2015); Kang et al. (2005); Solomon et al (2005); Twamley et al. (2014)
Use of ACP	Advanced care plan	Baker et al. (2008); Mitchel et al. (2015); Tan et al. (2006); Toce et al. (2003)
Awareness & advocacy	Among PPC staff, families, policy makers and society at large	Dangel (2002); Fernandez et al. (2011); Hubble et al. (2009); Perilongo et al. (2012); Steele et al. (2008a); Stephenson (2000); Toce et al. (2003)
Bereavement care	Structured guidelines	Ashby (1991); Bergstraesser et al. (2013); Corr et al. (1985); Danvers (2003); Davies et al. (2002); Di Gallo et al. (2006); Eggly et al. (2011); Epelman (2012); Jones et al. (2014); Lee et al. (2008); Kang et al. (2005); Rushton (2006); Schmidt (2011)
Spiritual care	Specific guidelines	Davies et al. (2002); Foster et al. (2012)
Research	Pain and symptom management	Ashby (1991); Downing et al. (2015); Shelton et al. (2011); Steele et al. (2008b)
	Objective criteria to identify patients and families with PPC needs (referral paths)	Bergstraesser (2013)
	Parents' and physicians' perceptions	Steele et al. (2008b); Hubble (2009); Toce et al. (2003)
	Collaboration and communication among team members	Mandac et al. (2014); Remke et al (2012); Rushton (2006)

Discussion

In light of the increasing number of children with life-limiting and life-threatening conditions, national and international organizations have developed a holistic and integrative model to care for this group and their families, as well as to provide guidance for healthcare professionals. Although PPC is gaining momentum, there is still an important gap between the guidelines and their implementation into medical practice. Our literature review identified strategies on how to overcome this gap so that all children who are eligible for PPC services might receive them. From the 54 studies included in the review, we found that most have looked at this problem mainly from a policy, clinical, or societal point of view, while only a few have examined the practical and conceptual pitfalls inherent to the guidelines. Our review suggests that some PPC principles are not always easily translated within a medical setting – they are seen as too theoretical to be able to deal with the complexity of the different stages of illness.

Some Trends throughout the Literature

Upon exploring the development of guidelines, we have found several trends that highlight both the emerging and continuing challenges faced by PPC over the last six decades. First, the very limited number of studies before 1998 is an indication of the slow growth of PPC. At the same time, the literature from this time period expressed an increased interest within the medical field for practice guidelines that inform staff and parents about the management of chronically ill and dying children. From 2000 onward, there is a growing recognition of the important role that guidelines play in the further diffusion of PPC. This is testified to by the steady increase in the number of publications. With the development of specific PPC guidelines, the main concern is the lack of palliative care services due to the absence of

adequate policy support (such as health insurance or national strategies that permit broad access). Attention is further placed upon the differences in availability of PPC services both across and within countries. In the studies published from the late 2000s onward, however, his concern takes a different form. There is an uncomfortable awareness that, despite increased policy support and available services, PPC is not readily accessible to all the children who could benefit from them.

Second, the inadequate implementation of PPC guidelines is mainly attributed to shortcomings within clinical practice and a lack of empirical research on PPC. This explains why recommendations concentrate primarily on educational and training programs for healthcare staff (including critical self-reflection), on the importance of multidisciplinarity (in particular, on the integration of PPC specialists into the primary care team to sustain the staff and guarantee seamless care without brusque transitions), and on the need for more evidence based research on the benefits of PPC. The latter concern is manifested in the increase in empirical research on the perceptions of parents and caregivers with regard to PPC from 2003 onward.

Third, the publications in the 2000s became more careful in respecting a clear conceptual distinction between hospice and palliative care. Although the hospice movement has been important to break the veil of silence surrounding death, these papers emphasize that this type of care should be redefined in function of the challenges posed by pediatric healthcare. They also underline that hospice care, due to its association with death, is still rife with stigma. Therefore, in line with the guidelines and motivated by the prognostic uncertainty within pediatrics and the family's improved quality of life, they embrace the paradigm shift within palliative care from a strict dichotomous to an integrative approach.

Finally, among the 54 included articles, 21 focus on end-of-life issues; however, only 4 of them date from the period between 2010 and 2015. Two of these four studies focus on

bereavement care in relation to the family and caregivers, and thus not directly on the death of the patients. This indicates that, for a long time, death and dying were considered to be central aspects of palliative care, whereas this connection has become less important recently. This finding is in line with those of other studies have highlighted the strained relationship between palliative care and the reality of death[82-84].

Some Missing Links and Prospects for the Future

Despite being a core principle of PPC, only a small number of publications report on the absence of clear guidance in PPC documents regarding bereavement care, the challenges of multidisciplinary care teams (e.g., hierarchy, competing values, communication), and training methods. More research needs to be done on bereavement care pathways[85] and effective teamwork. Also, further study is needed of effective teaching and training methods in regard to palliative care[86].

Another difficulty is linked to providing palliative care "from diagnosis onward." Although considered necessary to promote the acceptance of PPC, within medical practice it is often seen as unfeasible. Tools that can help physicians identify children and families with palliative care needs can be a way to address this concern and advance PPC. Some of these referral instruments are already under development[87,88].

Closely connected to the previous point, although PPC is not only meant for imminently dying patients, the question is whether the pendulum has not swung too far from end-of-life issues[82]. Palliative care has increasingly profiled itself as a more holistic medicine and as an alternative to the technical dehumanized medical tradition. This overall emphasis on quality of life and matters concerning the living might have contributed to the conceptual confusion around palliative care among both physicians and the lay public[82].

Having considered all of this, a critical assessment of both the research guidelines and medical practice is indeed needed in order to improve the implementation and outcomes of PPC.

Limitations

For this systematic literature review, a total of 54 studies were assessed. Our results should be interpreted with caution, as articles in languages other than English and German have remained unexplored. Some studies relevant to the issue might have been overlooked as a result of the search terms chosen. Aside from these limitations, ours is one of the first studies to explore barriers and recommendations for proper implementation of palliative care from the perspective of PPC guidelines in order to overcome the gap between recommendations and practice.

Conclusions

The evolution of guidelines in PPC confronts us with a true paradox: while in the 1980s and 1990s there was an increasing demand on the part of healthcare professionals to develop clear palliative care standards that would take into account the uniqueness of the pediatric population, two decades later there is a reluctance on the part of healthcare professionals to implement the model that was specifically developed in function of that population. Most studies address either the clinical gap or research gap to address this paradox. Accordingly, the recommendations found in a wide range of articles are related to the need to focus on training, education, and evidence-based guidelines. Future studies should continue to pursue empirical research on PPC and foster comprehensive educational and implementation programs that would make PPC an inextricable part of pediatric medicine. Aside from research on the perceptions of caregivers and parents about PPC, more research is needed

from the child's perspective in order to gain better insight into their needs and preferences, and thus enhance PPC services. With some important exceptions[89], the child's perspective on PPC is missing. Finally, to better align guidelines and medical practice, studies should begin to focus on the conceptual and practical shortcomings of published PPC guidelines.

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Chapter 8

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The Need for a Shared Understanding: Domains of Care and Composition of Team in Pediatric Palliative Care Guidelines

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Abstract

Conceptual confusion is a primary barrier to providing quality palliative care. This study aims to analyze pediatric palliative care guidelines from a conceptual perspective to facilitate a shared understanding of palliative care in pediatrics.

Five online data bases were searched systematically, in addition to a Google search. Analysis focused on the language used to determine the domains of pediatric palliative care and on the composition of the pediatric palliative care team.

Guidelines express consensus on four core domains: physical, psychological, social and spiritual care. However, conceptual vagueness exists with respect to the latter three as terminology is used inconsistently both within and across guidelines.

An inconsistent use of terminology affects the quality of pediatric palliative care nursing in various ways. Therefore, a shared understanding and unambiguous language must be envisaged. Furthermore, although guidelines agree on the most prominent team members they do not clearly indicate how these occupational groups should collaborate.

Introduction

Pediatric palliative care (PPC) seeks to care for pediatric patients, their families, and other significant persons using an interdisciplinary and holistic approach that focuses on several domains of care, such as psychological or spiritual care. Reduced child mortality rates and improved survival rates of pediatric patients with life-threatening diseases mean that there are more children who will need to access palliative care (PC), but only a fraction of these children actually receives PC [1]. Several barriers to implementation and sufficient provision of PPC have been identified, such as, organizational and economic obstacles[1].

Various scholars have also focused on the problem of conceptual confusion and in particular, on the lack of a shared understanding of PC[2-6]. They argue that good PC provision necessitates such a common understanding. Unfortunately, PC is often misunderstood both inside and outside the professional health care setting[4].

Recent studies in the US, Northern Ireland, the UK, Bangladesh, and Canada indicate that PC is relatively unknown among parts of the public and that persons who are aware of it often have a mistaken idea of its nature[7-11]. This knowledge gap may result from the ambiguous terminology used by health care providers[2]. Studies show that many physicians and nurses still equate PC with hospice care or end-of-life care[7], and those who know the difference, associate PC primarily with death and dying[12]. Another study shows that practitioners are often uncertain about how to translate PC in practice[13]. These misconceptions not only impact the lay understanding of PC, but also affect clinical practice in various ways, for example, whether and when patients are referred to PC.

Hence, using the right terminology is crucial as it can influence both medical practice (e.g. reduced number of late or non-referrals), and policy-making (e.g. allocation of more resources as a result of awareness). The same vision is shared by the European Association

for PC (EAPC): “it is obvious that an effective European approach to quality PC demands an unambiguous use of terms, which implies, as a prerequisite, the mutual agreement on the definitions of these terms”[3,p.280]. However, such a shared language requires a common standard of care to refer to[5]. With respect to PPC, this means that international guidelines that determine clinical practice need to be unequivocal. Knowing the concept of PC and being able to demarcate it from hospice care is insufficient if guidelines remain ambiguous.

Unlike the studies discussed above, this study’s rationale is to explore the conceptual consistency within and across international PPC guidelines. The focus was set on PPC guidelines because of the increasing number of children with life-threatening diseases and consequently, the increase in children who need access to PC. Furthermore, the study concentrates, in particular on two main principles of PPC: holism (the various domains of PPC care) and multidisciplinarity (the composition of the PPC team).

Consequently, our analysis covers two main research questions: (a) the functions, namely which domains of care are part of the palliative approach in the pediatric setting; and (b) the composition of the PPC team, namely which professions are part of it. In particular, this article will analyze the language used in PPC guidelines by focusing on the definitions of the PPC domains and on the PPC team composition in order to identify both conceptual consensus and possible conceptual inconsistencies. Implications of inconsistencies for clinical practice in general and PPC nursing in particular will be discussed. Since pediatric nurses often spend significantly more time providing care for the child and the family than other members of the PPC team, the impact of guidelines’ conceptual confusion on clinical practice is highly relevant for them. An enhanced understanding of PPC will enable pediatric nurses to better identify, understand and meet the needs and wishes of children with life-threatening diseases and to become more aware of their own crucial role within the PPC team

care[14]. The purpose of this analysis is to advance the development of such a clear PPC language to result in a shared understanding and, eventually, improved practice.

Methods

Research Design

A systematic literature search on both international and national guidelines on PPC was undertaken, resulting in 11 included documents. Subsequently, analysis of (a) guidelines' language used to determine the core domains of PPC and (b) the composition of the PPC team was carried out, thereby addressing two PPC principles: multidisciplinarity and holism.

Inclusion Criteria

The following inclusion criteria were used: documents have to be a) on PPC (infants, children, and adolescents are subsequently referred to collectively as "children"), b) developed by a national or international PC organization or a national agency, c) normative in the sense of providing standards regarding PPC, and d) written in English or German.

Exclusion Criteria

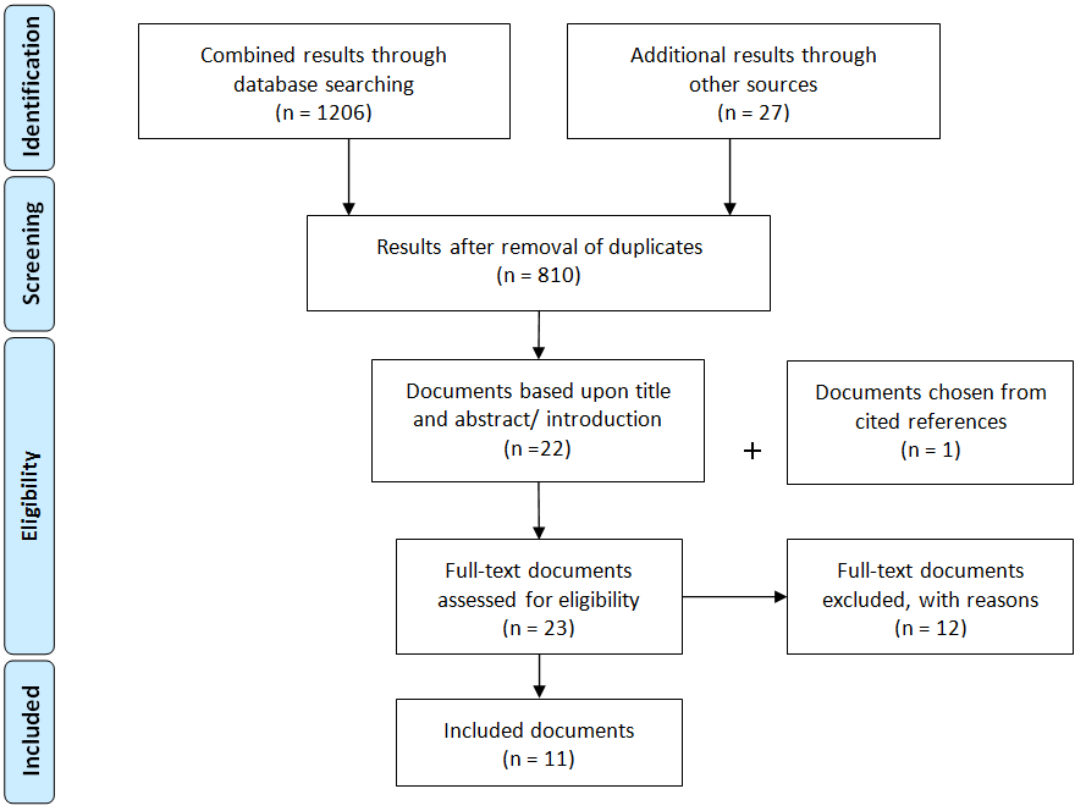
Articles on PPC guidelines were excluded and analysed in a systematic literature review[5]. Further, guidelines dealing exclusively with one aspect of PPC (e.g. spiritual care) or addressing only one group of children (e.g. neonates) were excluded since the aim was to identify the core domains of PPC in general. Guidelines that focus exclusively on one group or one aspect naturally do not touch on the question which set of domains constitutes PPC.

Search Strategy

The literature search was comprised of two parts. First, employing the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) framework[15], five online databases (Scopus, PubMed, PsycInfo, Web of Science, and CINAHL) were scanned,

combining search terms through Boolean algebra as follows: (Pediatric* or child* or adolescent*) AND (palliati* or palliative care or hospice care) AND (guidelines or recommendations). Second, a Google search was next performed using the above-mentioned search terms expanded by the inclusion of the term "guide" in the last parentheses: (guidelines or recommendations or guide). Scanning the five data bases resulted in 1206 documents, and the Google search in 27. Subsequently, 413 out of 1206 (databases) and 10 out of 27 (Google search) were identified as duplicates; 810 documents remained (Figure 1).

Figure 1. Search process using PRISMA Systematic Review of Literature



During the next phase, two researchers screened all 810 titles and abstracts (or introductions, respectively), resulting in 22 documents that were potentially eligible. The references of the latter were checked for additional documents. Through this process 1 document was added. In a final step, the first author read the full text of the resulting of 23 documents. Evaluating

these documents led to the exclusion of 12 documents because they (1) were not the latest version of a document, (2) focused mainly on adult PC and touched only superficially on PPC, (3) addressed only one sub-group of children, or (4) presented exclusively facts without suggestions on best practice. A final set of 11 guidelines published between 1998 and 2013 fulfilled the inclusion criteria (table 1).

Analysis

The analysis was carried out in several steps. First, all domains and team members were extracted by reading one document at the time. The focus was set on how guidelines defined or outlined PPC, in particular which domains of care were considered constitutive for PPC, and on who was considered a member of the PPC team. Thereby, any domain of care that was used to define PPC was included in a list of all possible domains and any occupational group that was part of the PPC team was included in a list of all possible team members. Out of the eleven documents, 10 documents explicitly referred to domains of care that constitute PPC and eight to which occupational groups constitute the PPC team.

Second, those domains that were used in all documents to define PPC were identified as core domains. For example, psychological care was considered a core domain because all guidelines referred to it when defining PPC. Furthermore, all occupational groups were ranked according to how often they were considered a part of the PPC team by the guidelines (list in a descending order). This step resulted in 4 core domains of PPC, namely, physical, psychological, social, and spiritual care and in a descending list of how often a specific occupational group was considered a member of the PPC team along with information on which documents referred to the respective occupational group.

Third, both authors re-read and examined the guidelines, focusing on the comparison of the guidelines' terminology with respect to the four core domains. This procedure allowed

determination of both consensus and inconsistencies across and within guidelines. In particular, analysis of the guidelines' language used to designate domains of PPC employed the following categories of how two terms can interrelate: (a) distinction on different levels, (b) distinction on the same level, (c) used interchangeably, or (d) merged to one domain.

The term "distinguished on different levels" refers to a subordination of one domain to another, for example emotional needs are subordinated to psychological needs. The term "on the same level" points to two independent domains without one being subordinated to the other, for example psychological and physical needs are two independent core domains. "Interchangeably" means two terms are used as synonyms to designate the same domain of care, like for example social and psychosocial. A "merged domain" is characterized by two terms that are used jointly (at the same time), for example psychological-emotional care.

Results

In total, 11 documents were analyzed, 3 from internationally recognized organizations (1 from the World Health Organization, 2 from the EAPC Taskforce for PC in Children), 8 from organizations working on a national level, spanning seven North-American or European countries (USA, UK, Canada, Ireland, Scotland, Germany, Austria).

Table 1. List of included guidelines

Document	Self-description and purpose
WHO (1998)	Guide to pain management in childhood cancer and PC in children [16]
CHPCA (2006)	Guide for standards of practice, service delivery, program and policies [17]
IMPaCCT (2007)	Defining and identifying standards of care [18]
NHaPCO (2009)	Guide for palliative and/or hospice programs providing care [19]
EAPC (2009)	Examination of the state-of-the-art and the need for PPC [1]
DoHC (2009)	Foundation for developing children's PC services [20]
ACT (2009)	Guide to the development of children's PC services [21]
SCYPPEx (2012)	Framework for the delivery of PC for children and young people [22]
DHPV (2013)	Principles for hospice and PC for children and adolescents [23]
BfG (2013)	Concept for hospice and PC for children [24]
AAP (2013)	Statement with principles for PPC [25]

Note. AAP = American Academy of Pediatrics; ACT = Association for Children's Palliative Care; BfG – Bundesministerium für Gesundheit, Austria; CHPCA = Canadian Hospice Palliative Care Association; DHPV = Deutscher Hospiz- und PalliativVerband e.V., Germany; DoHC = Department of Health & Children, Ireland; EAPC = European Association for Palliative Care; IMPaCCT = International Meeting for Palliative Care in Children; NHaPCO = National Hospice and Palliative Care Organization, USA; PC = Palliative care; SCYPPEx = Scottish Children and Young People's Palliative Care Executive Group; WHO = World Health Organization.

Core Domains of Pediatric Palliative Care

All documents, except for one that does not list particular domains of care[23], identify four core domains, namely physical, psychological, social and spiritual care. These domains are further discussed below and exemplified through quotes (table 2). Besides these four core domains, other domains were used to define PPC. However, none of these secondary domains was used by all guidelines. Practical care, which refers to activities of daily living and home-based services, is considered a separate domain in three documents[17,22,25]. Other aspects of

PPC, such as loss, grief, bereavement, end-of-life care[17], cultural care[24], or developmental care[17,19] are rarely considered separate domains of PPC. Analysis of language revealed that conceptual vagueness exists especially with regard to psychological, social, and spiritual care due to inconsistent terminology both within and across guidelines (table 2).

Table 2. Examples of conceptual vagueness

Domain	Category	Examples
Psycho-logical vs. Emotional	Distinction: same level	*"The PC team (…) address[es] the physical, psychological, emotional (…) needs (…)."*[18]
	Distinction: diff. levels	*"Psychological needs: (…) continual emotional support to help the child."*[1]
	Interchangeably	*"Physical, emotional, and spiritual suffering"* *"Physical, psychological and spiritual comfort"*[19]
	Merging	*"Psychological-emotional needs"*[24]
Social vs. Psychosocial	Distinction: same level	*Separate domains: "psychosocial care" and "social care"*[17]
	Interchangeably	*"(…) minimize the child's physical, psychosocial, and spiritual (…) suffering"* *"(…) address physical, social (…) and spiritual needs of children"*[19]
Spiritual vs. Existential	Distinction: diff. levels	*"Spiritual care: (…) Formal caregivers address the child's and family's existential questions (…)."*[17]
	Merging	*"Relieve suffering across multiple realms, including (…) existential or spiritual (why is this happening?)"*[25]
Spiritual vs. Religious	Distinction: same level	*"Formal caregivers determine what distinguishes spiritual from religious practice"*[17]
	Distinction: diff. levels	*"Spiritual needs: (…) religious background."*[1]
	Merging	*"Spiritual and/or religious care" or "spiritual/religious"*[18]

Note. Diff. = Different. PC = Palliative care; PPC = Pediatric palliative care.

Physical care is mostly identified with pain. Pain is used to indicate not only physical, but also psychosocial, spiritual[19], and emotional aspects of pain[16]. Frequently, the physical and emotional aspects of pain are grouped into broader categories of symptom management or pain management. Finally, one document uses the terms "clinical needs" and "physical needs" interchangeably when referring to children's needs that require physical care[1].

Psychological care is often used inconsistently across documents because the concept is not sufficiently demarcated from emotional care (table 2). First, some documents distinguish these two as different concepts. They either clearly distinguish psychological needs from emotional ones on the same level of definition[18], or they consider emotions to be a subcategory (different level of definition) of the broader psychological domain[1,19]. Second, one of the documents only uses the term emotional care and does not mention psychological care when referring to this particular need of a child[22]. Third, in one case the two terms are merged into a single domain of psychological-emotional care[24]. Conceptual inconsistency can be found not only across, but also within one and the same document. For example, two documents list emotional care as a PPC domain, thereby either implying that it covers psychological care or that psychological care is not a domain of PPC, but then clearly distinguish psychological from emotional support elsewhere, thereby apparently referring to different concepts [18,20]. Furthermore, one document lists both psychological well-being and the emotional impact of an illness as two separate subcategories of PPC's domain of psychosocial care, thereby distinguishing emotional and psychological on the same level and subordinating both terms to psychosocial care[17]. Finally, in some cases both terms are used interchangeably[19,25], for example, one document first lists realms of a child's suffering as follows: physical, psychological, practical, and spiritual, before it later enumerates "physical, (…), emotional, practical and spiritual needs of the child", thereby apparently equating emotional and psychological needs across the two enumerations[25,p.968].

Social care is another concept that is used inconsistently both across, but especially within documents because it is not sufficiently demarcated from psychosocial care (table 2). One document uses the terms "social care" and "psychosocial care" to indicate two separate domains by providing a separate section for each of them[17], thereby distinguishing them on

the same level. Most guidelines, however, only use social care to designate this particular core domain of PPC[1,16-22,24,25]. In some cases, the term is coherently used throughout the text, hence avoiding conceptual vagueness[1,18,22], but in others it was used interchangeably with psychosocial care[16,19,20,24,25]. For example, one document first refers to the social domain (besides the psychological, spiritual and physical) that is addressed by PPC and later states that "psychosocial (…) domains of distress" or "psychosocial (…) needs" have to be addressed (besides physical, emotional, practical, and spiritual needs), thereby using both terms to describe the same domain and *a fortiori* equating both terms[25]. Another one uses the two terms interchangeably across two analogous enumerations of care domains (table 2); the document uses "psychosocial" in the first enumeration, and the term "social" in the second[19]. Moreover, one document uses the psychosocial domain to define PC in general, but the social domain to define PPC, indicating that this represents a conceptual difference either between social and psychosocial or between PC for children (this particular PC includes the social domain) and adults (this particular PC includes the psychosocial domain)[20].

Spiritual care is frequently mentioned in connection with existential care (table 2). One document distinguishes the two concepts on different levels by subordinating existential to spiritual care[17]. A few guidelines only use the term spiritual care, thereby avoiding conceptual vagueness[19,24]. Finally, one document lists spiritual care as a separate domain in the headings, but makes references to "spiritual/existential" or "existential or spiritual" throughout the text [25], thereby creating a merged domain. Other documents set spiritual care alongside religious care (table 2). One document mentions the "religious background" as an aspect of spiritual needs, thereby subordinating religious to spiritual care[1]. Furthermore, two documents differentiate spiritual and religious care on the same level, one as separate parts of an end-of-life plan[20], the other one requires formal caregivers to distinguish spiritual from

religious practice[17]. Finally, both terms are merged to create one domain: for example, "access to spiritual and/or religious care" and "spiritual/religious worker"[18].

Composition of PPC Team

With the exception of three guidelines[19,22,23], all other documents discuss the particular multiprofessional composition of the PPC team and insist on its interdisciplinary collaboration. The most frequently listed members are: physicians and nurses (all 8 documents), followed by social workers who were not listed in one document[16], psychologists[1,17,18,21,24], chaplains[16,17,21,24,25], volunteers[16,17,21,24,25] (5 documents), and physiotherapists[1,20,21,24] (4 documents). Three documents listed occupational therapists[1,20,21]. Child-life therapists[17,18] and pharmacists[17,21] are mentioned in 2 documents; spiritual advisors in 1[18]. All documents require the PPC team to collaborate with the family and the child. Several documents highlight the importance of a designated person who coordinates the PPC services, such as a care coordinator[18,19,24], key worker[18], navigator[17], lead doctor and nurse[22]. However, no clear indications are given on how the team members (how, when, under what circumstances, authorities, procedures etc.) can or should collaborate.

Discussion

In the analyzed documents, there is a broad consensus on the set of four core domains that constitute the holistic approach of PPC. However, across and even within some of the documents three domains lack conceptual clarity, as the use of the terms is often inconsistent. Since an effective approach to quality PC necessitates an agreement on definitions of terms, it is paramount to examine how this conceptual confusion might affect the quality of PPC.

Pediatric Palliative Core Domains

First, it remains unclear what separates psychological from emotional care, social from psychosocial care, and spiritual from religious or existential care. Whereas the term "psychological" embraces both cognitive and emotional aspects, the term "emotional" refers to affective states (e.g. pleasant, unpleasant) and thus has a narrower focus than psychological care[26,27]. An enhanced and broadened focus would better respect a child's right to development (enshrined in article 6 of the United Nations Convention on the Rights of the Child (UNCRC) which, among other factors, requires support for intellectual development[28,29]. For the PPC context, intellectual needs are cognitive aspects of care, such as those related to a child's growth in knowledge, critical thinking, learning new things, or making sense of the illness experience. These aspects reach beyond mere schooling and education, which represent rather formalized areas of children's intellectual development, and need to be made available. Apart from neglecting the cognitive needs of children, an overly narrow focus on emotional needs might reinforce the assumption that children, due to their age and developmental stage, are cognitively impaired, lack decisional capacity, and need surrogate decision-makers. This reinforcement decreases the likelihood of children's involvement in the decision-making process, for example, on whether to start PC. Including

children in these decisions is unanimously recommended by all guidelines, as well as by the UNCRC[29]. Therefore, the term psychological is preferable as it encourages health care providers to involve children, simultaneously identifying and meeting both cognitive and emotional needs.

Furthermore, not sufficiently demarcating social from psychosocial care leaves the question open of how these types of care are different. Unlike "social," the term "psychosocial" pertains to the interwoven psychological and social aspects of care as well as to the social determinants of health[30]. In contrast, social care more narrowly focuses on the social relations and significant others themselves (e.g. family, friends). However, social relations sui generis have a direct impact on the psychological state of an individual, and therefore, the psychological dimension is always implicitly considered by social care. That being said, one can legitimately raise the question what the term "psychosocial care" actually adds to social care? Ultimately, the term "psychosocial" more directly refers to the twofold meaning of social relations, namely the relations themselves and their impact on the psychological state. Given this blurred demarcation, it is unsurprising that studies have shown that the provision of psychosocial care is hampered due to unclear responsibilities among team members[31]. Especially with respect to psychosocial care, it is therefore important to not just articulate that team members work together, but to critically examine the collaboration[31].

Finally, spiritual care lacks conceptual clarity as it is narrowly connected with existential or religious care, but the relations among these terms remains blurry. Given the subordination of existential and religious care to spiritual care, spiritual care appears to be the most suitable term in order to designate this particular core domain. This is further backed up by experts' understanding of spirituality as composed of various elements, such as religious and existential aspects[32]. Using spiritual care as the most inclusive term might also align with modern pluralistic societies and suggests a neutral, inclusive stance of medical guidelines.

Second, diverging definitions of psychological, spiritual, and social care might hinder an adequate understanding of the core domains of PPC, and this may influence the creation and coordination of an effective PPC team. The members of the team can only reach an agreement on how to operate if they have a clear understanding of their roles and responsibilities. Misunderstandings about each other's tasks, skills and expertise might cause interpersonal conflicts and competition[33]. For example, a study has shown that with respect to psychosocial care, PC team members exhibit a lack of clear role boundaries. Some members believe that any team member can meet the patient's psychosocial needs[31]. Interestingly, nonspecialist psychosocial team members perceived these unclear roles as positive, specialist psychosocial team members as negative. This situation causes a division of the team as it leads to so-called "contested realms", that is, team members attribute specialist expertise to themselves while doubting the expertise of colleagues[31].

Due to their nature as multiskilled professionals, pediatric nurses are especially prone to facing overlapping roles and responsibilities when working in a multidisciplinary team. Again, a shared understanding of language that avoids ambiguous terms and a clear understanding of individual roles facilitate skillful communication within the team. Members of the team, and nurses in particular, need to be able to articulate their expertise and knowledge in order to maximize the benefit from the numerous skills of the team members[33].

Literature also shows that reflective practice among team members, for example hearing narratives of experiences by other members, can additionally improve teamwork[34]. However, in order to improve practice in such a way, it is necessary that the team members share the same terminology and use it consistently.

In an efficient team that works successfully towards its goals, every member needs to have a clear understanding of their own contribution to the team[35]. Besides interpersonal conflicts and competition, differing uses of terms among team members can result in partially

conflicting understandings of one owns contribution, and thereby in different types of care, in involving different experts, and possibly in not meeting a child's needs sufficiently.

The term psychosocial care indicates the need to focus on the impact that social relations have on the child's psychological state, for example through psychotherapy or psychiatric drugs. Social care, puts more emphasis on the social relations themselves that can be addressed by a social worker through involving significant others within a systems approach. Even though psychological care refers to both a child's intellectual needs, for example explaining the cause of the disease in a developmentally appropriate way, and emotional needs, for example dealing with despair with the help of a psychooncologist, the guidelines sometimes represent only the emotional aspect and neglect the intellectual one. Finally, if spiritual care is limited to providing religious care, a chaplain might be the best choice, but if it rather embraces existential care, an occupational therapist, which helps to make hand prints for reminiscence is better meeting the requirements. As apparent from the preceding examples, the particular understanding of one core domain determines not only which occupational group is involved, but also the factual care outcome.

Composition of Team

First, all analyzed documents emphasize that PPC should be put into practice by a multiprofessional team whose work is not limited to the hospital setting, but that works across several health care settings and adapts care dependent on a child's particular needs. The team members should collaborate in an interdisciplinary way.

Results of the analysis revealed that the core of the hospital's PPC team is comprised of a physician and a nurse. Social workers represent the second most-mentioned group; psychologists, clergy, and volunteers the third. Other staff members, for example pharmacists, are listed only occasionally. The results confirm findings of other studies which

show that for PC experts the core team is composed by physicians and nurses (absolute minimum), psychologists, social workers, and physiotherapists[36]. However, in the same study controversies were found regarding psychologists and chaplains which are somehow mirrored by the fact that some of the analyzed documents do not list these groups.

Second, the question of how the team members' interdisciplinary collaboration should look like was not addressed substantially, as this best practice advice was only mentioned but not elaborated in depth. This is unfortunate, since, according to Remke and Schermer, a shared vision of how the team's objectives will be achieved promotes successful team work by increasing the degree of efficiency, of trusting one another, and of satisfaction with the own role[37]. Besides, it is vital for pediatric nurses not only to know the team members, but to be provided with basic information on the interdisciplinary approach[14].

The disagreements regarding the final team composition and the unspecified interdisciplinary collaboration represent a double-edged sword. On the one hand, this unfixed model takes account of the necessity of a flexible, individual-based, and context-sensitive approach of PPC by preserving open-endedness and adaptability of care[38]. On the other hand, it opens the doors to interpersonal conflicts in light of team members' tendency to protect their own expertise in case of overlapping skills[33].

Third, most guidelines agreed on designating a coordinating person that serves as both a port of call for the family and, at the same time, as a centre of convergence for important care-related issues. Recent studies emphasize that coordination of a multiprofessional teamwork is crucial for providing quality PC and reaches beyond only one single coordinating person, for example to multidisciplinary team meetings or team training programs[31,35]. However, one designated coordinating person seems to conform to a minimum of coordination which has to be at hand in every team at every moment. A coordinating person should address the task of

monitoring the team composition and collaboration, thereby helping to avoid interpersonal conflicts among team members and unmet needs of the child.

Because pediatric nurses are key team members who provide multiple domains of care and spend significant amounts of time with the child and family, they represent a suitable candidate for coordinating care that is in the best interest of the child. In fact, a study has shown that referral rates were greater in PPC teams with an advanced nurse practitioner[39].

Fourth, the two analyses (domains and team composition) converge because the four identified core domains of PPC can be covered by those occupational groups which are considered members of a PPC team by the majority of the guidelines.

Limitations

First, only guidelines that were written in English or German were included. Second, important guidelines might have been overlooked because of the search terms that were chosen for the literature search. Third, to some extent linguistic differences reflect cultural and historical differences which may have an impact on the concrete form of PC and on terminology. Consequently, differences in terminology will continue to exist and are legitimate, but this study's findings rather reflect differences due to conceptual confusion and a not sufficiently attentive use of terminology.

Conclusion

Our analysis indicates a broad consensus on four core domains. At the same time it reveals a lack of conceptual clarity for three domains. This is problematic insofar as conceptual clarity is an important prerequisite for quality PPC. Consequently, these terms need clarification, whereby the avoidance of using multiple terms for designating the same domain can serve as

a first step. The terms should be used more deliberately, considering (seemingly) small linguistic differences and their (likely) effects on clinical practice.

Since pediatric nurses are involved in all core domains of PPC, an unambiguous terminology that facilitates quality PPC is especially beneficial for them. Despite the benefits of shared terminology, the authors acknowledge that several barriers complicate a consensus. With respect to terminology countries differ considerably regarding history and language. Similarly, a study on understandings of PC, highlights the different historical developments of PC among European countries as potentially hindering[36], and considers the specific nature of PC as set out by the WHO to be a barrier to reaching an agreement on the scope of PC. Still, any PC definition should be based on patient needs and corresponding domains of care.

Apart from affecting medical practice, conceptual vagueness might magnify the already existing knowledge gap of PC among the lay public. This pathway is mediated by the inconsistent use of terminology among professionals. Inconsistent guidelines lead to an inconsistent use of terminology among health care professionals which eventually contributes to laypeople's knowledge gap and confusion; Bergstraesser has rightly stressed this interplay[2]. It is self-evident that greater knowledge, less confusion, and raised awareness for PPC on the part of the lay public (but also within academia and clinical practice) promotes higher acceptance and consequently, in the long run, more resources allocated to PC.

With respect to team composition, guidelines agree on a standard that contains physicians and nurses, and that is complemented by several other professions. The guidelines neither specify how these occupational groups should collaborate nor who should be part of the complementary group. This situation comes along with benefits and risks. It is of crucial importance to work toward further improvement, for example by designating a coordinator who utilizes this ambiguity to tailor PC to a child's specific needs.

Finally, the authors acknowledge that PPC must be an individually tailored endeavor that cannot be fully formalized. Aiming for a shared understanding of domains in order to facilitate optimal care does not contradict this notion. On the contrary, quality care requires pre-existing structures and expertise on which the team can build best possible care, thereby meeting a child's needs by adapting to the particular illness profile[6]. This study contends that the quality of PC can be facilitated by a consistent use of language, as expressed by R.W. Emerson: "thought is the bud, language is the blossom, and action the fruit behind it".

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Chapter 9

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Is Decision-Making Capacity an "Essentially Contested" Concept in Pediatrics?

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Summary

Decision-making capacity (DMC) is the sine qua non of informed consent, since the latter is only valid if it is provided voluntarily, knowingly, and rationally[1]. This, in turn, requires a patient's DMC to understand, appreciate, and use the information that was disclosed[2]. Further, to become legally binding a competent patient's decision necessitates DMC. Most legislations presume a patient to be competent from the age of 18 onwards[1,2]. However, applying the concept of DMC in the pediatric setting entails various challenges[3]. One main reason is the persisting view of children as a cognitively impaired class that lacks decisional capacity[1,4]. As a consequence, in clinical practice parents mediate the (minor) patient-physician relationship as surrogate decision-makers resulting in a triadic constellation of shared decision-making[3,5]. Having considered this, the question arises whether such an understanding of capacity that mainly refers back to adult-centred notions (e.g. autonomy) can facilitate children's involvement in decision-making.

Conceptual and Definitory Challenges

Despite lacking legal competence, children are increasingly involved in decision-making as a result of ethical guidelines' recommendations[6] and, at the same time, are generally less perceived as impaired adults but rather as active beings that are part of the society[7]. Nevertheless, adequate implementation of children's involvement still proves to be difficult because of numerous barriers[6,8]. From a conceptual perspective it remains unclear what is meant by "having DMC". There are two main conceptualizations of DMC. On the one hand there is the mainstream account according to which DMC should be assessed based on the process regardless of the decision outcome[9]. Here, DMC is conceptualized procedurally as a mental ability determined by the principles of logic and rationality[10]. Theorizing DMC as a process of rationality is strongly linked to notions of autonomy and informed consent which

perceive persons as rational agents. On the other hand several criticism of such a cognitivist model of capacity prepared the ground for alternative models[11-14]. The latter share the conviction that the former neglects the interdependence of the self and argue that other determinants of DMC need to be acknowledged (e.g. social, political). Further, they express the concern that a narrow cognitivist and rationalist conceptualization of DMC could lead to a discrimination of cognitively impaired persons (e.g. children). Therefore, they argue for considering so called substantive or non-cognitive factors (e.g. emotions, beliefs, values)[15]. Besides differing conceptualizations of children's DMC, various attempts to develop an empirical assessment tool have been made. However, these tests are based on the underlying definitions of and assumptions regarding DMC[16]. For example, Hein et al. have developed an instrument for children by modifying the MacArthur Competence Assessment Tool for clinical research[17,18]. The latter focuses on cognitive criteria for capacity as set out by Appelbaum and Grisso[19]. Again, this focus on cognition is normatively laden as it is derived from the ideal of an autonomous and self-directed adult[20].

Bearing in mind the different conceptualizations of DMC and their perpetuation into different definitions of DMC and, in the end, into various assessment tools for DMC, we propose to re-think the traditional cognitivist model by approaching DMC through the lens of Gallie's notion of *essential contestability*[21]. Therefore, in what follows, we outline the seven criteria of essential contestability according to Gallie as well as their applicability to DMC.

Essentially Contested Concept: Criteria and Applicability to Decision-Making Capacity

When the philosopher Walter Bryce Gallie introduced the notion of essentially contested concepts six decades ago[21,22], he was primarily concerned with the way concepts are used and applied within societal debates and not so much with the philosophical nature of concepts[23]. He defined essentially contested concepts as "concepts the proper use of which inevitably

involves endless disputes about their proper uses" that are unresolvable through the use of rational arguments[21,p 169]. It is important to note that this definition does not refer to conceptual confusion or to strongly disputed concepts, which represent rather practical problems and, in principle, are resolvable[24,25]. In contrast, a concept is essentially contestable if it inheres the potential to generate discussions that are somehow undecidable[26].

Gallie provided seven criteria for essentially contested concepts[21]. *Criterion I*: they have to be evaluative, that is they have to convey a value judgement. Assessing the capacity of a person always implicitly expresses a normative statement, namely whether a person *should* be allowed to make decisions. *Criterion II*: as they embrace various dimensions, they are internally complex. DMC contains a number of internal components, such as procedures, rational cognition, social and individual factors etc. *Criterion III*: Because of the second criterion they offer an immense number of conceptions. Depending on the focus of the respective approach, different conceptions of DMC exist. *Criterion IV*: they have to be open-ended and dynamic in nature. Besides the existence of various approaches to DMC, the openness criterion is also evident in the fact that the set of conceptions of DMC is changing over time, for example "reasonableness" was required for DMC[27]. Also, conceptions may be adapted in face of changed circumstances, as it is the case with the variable standard for DMC (e.g. greater levels of capacity for more complex decisions). The aforementioned concepts represent "the four most important necessary conditions to which any essentially contested concept must comply"[21,p 172]. There are three further criteria. *Criterion V*: essentially contested concepts are interpreted differently and used aggressively and defensively against other users' conceptions. Without any doubt, this is the case for DMC. *Criterion VI*: these concepts are exemplar that is they agree on a shared minimum[28]. The various approaches portray DMC as a gatekeeper to autonomy. *Criterion VII*: the ongoing

disputes about an essentially contested concept cause improved understanding and a fuller realization of these concepts. In the following, we will explore in how far this is fulfilled.

Conclusion: Usefulness for Decision-Making in Pediatrics

The current predominant perception of DMC as a cognitive ability residing within an individual obscures the multidimensional determination of capacity (relational, cultural, cognitive etc.) and suggests that capacity could objectively assessed. Thereby, it neglects the fact that capacity always evolves within a cultural framework which shapes that same capacity. Generally, understanding capacity as an essentially contested concept gives space for alternative approaches and emphasizes the questionability of the current mainstream model of DMC. Particularly, it enables to question the cognitivist standards that are underlying children's capacity assessments which not only shifts away from narrowly focusing on cognitive abilities when assessing DMC, at the same time it refers to the possibility to enhance capacity by taking into account extra-individual factors[29,30].

Since children are still seen as lacking cognitive abilities of "normal adults", they are not granted the assumption of having decisional capacity, but carry the burden of proof for capacity[31]. This is not to deny that children are still developing and lack certain cognitive abilities, but to underscore that children, to a great extent, are emotionally, socially, and financially dependent on others[32]. As a result, their capacity might be hindered by parental attitudes and physicians' experience, workload, and values.

In conclusion, non-cognitive determinants of children's DMC need to be considered. This requires a shift away from a deficit model of capacity (lacking cognitive abilities) to one of shared responsibility, where all parties involved in decision-making contribute to capacity[6,31]. Defining DMC as an essentially contested concept and, thereby, allowing alternative models does not inhibit, but facilitates such a shift.

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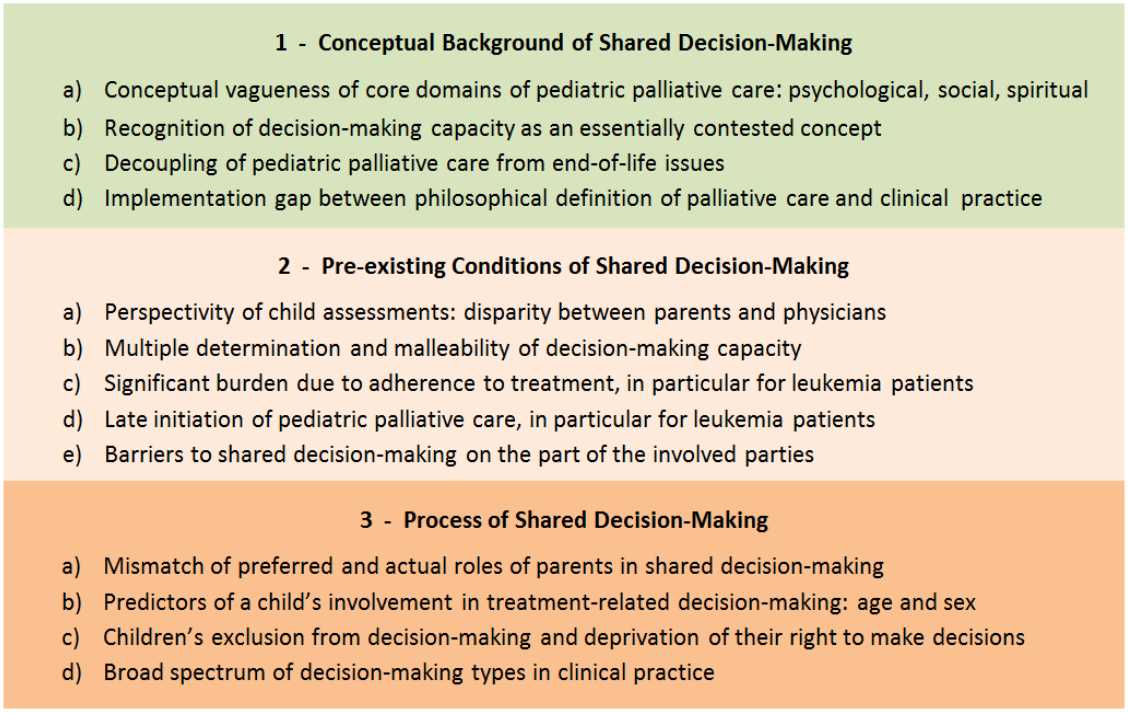
Chapter 10 – General Discussion

1 Integrating Empirical Results and Ethical Analysis

This section is arranged in three parts (figure 1). The first part presents results related to the conceptual background of SDM with a focus on the conceptualizations of PPC and decision-making capacity. The second part illuminates pre-existing conditions of SDM, such as perceptions of involved parties, evaluations of children's capacities, treatment-related burden, and barriers to the implementation of PPC. The third part discusses the process of SDM, namely a child's involvement, the parents' roles, and the structure of decision-making.

Figure 1. Synopsis of main findings



1.1 Conceptual Background of Shared Decision-Making

With respect to the core domains of PPC, the conceptual vagueness within and across guidelines (chapter 8) underscores the need for a shared understanding and unambiguous terminology. A mutual understanding of these terms is a prerequisite for the provision of

quality PPC[1]. As demonstrated, failure to achieve a shared understanding leads to interpersonal conflicts and competition within the PPC team, affects the factual care outcome, and colors the perception of children and their decisional capacity. For example, by narrowly focusing on emotional needs, cognitive needs are likely neglected, which might reinforce the perception of children as cognitively impaired and, in the end, increase the likelihood of their exclusion. As such, a consistent use of language not only helps to facilitate quality PPC and intra-team collaboration, but also improves children's involvement.

The proposed recognition of decision-making capacity as an essentially contested concept (chapter 9) overcomes the rationality-centered approach to capacity that originates from an individualistic understanding of autonomy. This is important, because solely reifying capacity as a cognitive ability of a child overshadows social, biographical, and cultural determinants. This is of course not to say that practitioners should refuse to assess capacity, but rather to point out that conceptualizations of capacity cannot be found or detected – as if capacity is a mere matter of fact, instead they are always selected, for example from within a given cultural framework[2]. Doing justice to the complexity of the concept of capacity as well as to children's actual capacities requires analysis beyond a determination solely on the basis of measurable cognitive properties[2]. Any ethically sound conceptualization of decision-making must analyze how capacity is determined. In doing so, the reported study (chapter 9) concluded with recognizing capacity as an essentially contested concept, thereby, allowing alternative approaches to inform and enrich the ongoing debate.

The reported findings on the domains of PPC (chapter 8) and on the conceptualization of decision-making capacity (chapter 9) have a point of convergence. The former warns against reinforcing the assumption of cognitively-impaired children and the latter rejects identifying capacity as an exclusively cognitive trait and, thus, de-emphasizes the significance of cognition for conceptualizing and assessing capacity. Interestingly, both neglecting and

overemphasizing cognitive aspects are likely to result in children's decreased involvement in SDM. Neither bypassing cognitive aspects nor exclusively considering them accurately assigns the value to children's cognitive capacities when thinking about their role in decision-making. Finally, the fact that cognitive aspects are sometimes neglected and sometimes overemphasized mirrors the difficulties to construe minors' decisional capacity.

Furthermore, an important insight into how the conceptualization of PPC has changed over the past decades is provided (chapter 7). Within more recent guidelines (2010-2015), the role of end-of-life issues has become less important, while the notion of quality of life has garnered more attention. This change may have contributed to the conceptual confusion between palliative and hospice care among the lay public and physicians, and it leads to the question, has the pendulum swung too far away from end-of-life issues[3]. Whilst turning away from an emphasis on end-of-life issues may help to sharpen the focus on quality of life, it is questionable whether this refocus actually represents the lived realities of families whose experiences, in face of cancer, often are characterized by contemplation of death and dying.

Related to the decoupling of PPC from death and dying, a final concern (chapter 5) is that although pediatric oncology providers are aware of the PPC principles outlined by the WHO, they insisted that it is best provided when the disease is unresponsive to curative treatment. How to implement such a philosophy into medical practice remains a lingering challenge[4]. Today, practitioners in Switzerland are confronted with a dilemma. Either they attempt to rebrand PPC, for example as comfort or supportive care, to counter families' feelings of abandonment and avoid associations with end-of-life, or they continue to use the term with its associated connotations. While the former loses sight of the original aim and risks to deny the significance of death and dying in PPC, the latter continues to make families uncertain and, thereby, impedes the acceptance of palliative care among the public. As long as this situation prevails, proper implementation of PPC and discussion of PPC within SDM are hindered.

1.2 Pre-existing Conditions of Shared Decision-Making

This thesis identified various conditions that already exist when SDM is initiated. First, reported findings reveal some characteristics of the parties involved in SDM. The disparity between parent and physician perspectives (chapter 2) indicates systematically different views of the child. Parents deemed their children more capable than physicians did regarding the child's understanding of the diagnosis and prognosis. Additionally, compared to physicians, parents evaluated the impact of the disease on their children as worse (e.g. more suffering, longer duration), and finally, parents believed their children to be more satisfied with decision-making than physicians did. Regardless of which outlook is closer to the truth, it should be acknowledged that these varying perceptions might negatively affect SDM due to a limited understanding of the other party's perspective or due to disputes about the child's capacities and health status. SDM, which requires parties to (a) share information, (b) work towards a consensus, and (c) reach an agreement[5,6], becomes an empty formula without serious consideration for how these parties differ from each other. This finding is important as it reminds everyone involved in SDM of the underlying perspectivity of what seem to be factual and intersubjectively shared (by physicians and parents) assessments. Whether they are shared cannot be assumed, but warrants active research.

The reported findings on how treatment comprehension and decision-making capacity are evaluated in children (chapter 2) support assessment of capacity based on the type of decision, among other factors[7]. While understanding diagnosis and the response to treatment were judged to be the easiest (turning points: 6.5 and 6.0 years), making treatment decisions was considered as more difficult (turning point: 11.5 years). Again, SDM always refers to a particular decision that determines whether (and to which extent) or not a child should be involved. In this respect, decisional capacity aims to align the decision's difficulty and subject with the child's characteristics. Similar to this, another study (chapter 3) shows that

most children older than 13 years of age were involved in decisions regarding palliative care (turning point: 13.5 years), suggesting that end-of-life decisions were evaluated as most difficult. Overall, findings on the evaluation of children's capacity (chapter 2 and 3) underline the necessity to think of capacity as changing across decisions.

Second, results of the reported studies reveal treatment-related variables that influence SDM. As described, children suffering from cancer and their families face a significant burden of treatment that goes beyond just side effects (chapter 4). Adherence to the treatment regime has highly disruptive effects on families' daily lives, for example on working lives, social lives, and leisure activities. This study's findings advocate for thinking about burden of treatment from a family perspective. Thus, health care professionals must be aware of the disruptive effects that pervade the entire family unit. Also, recognizing the family as one unit of care means to apply a systemic approach to SDM: to address individuals as members of a family unit, instead of each individual separately, and to be aware of the importance of the stability of family structure for the child. Finally, a high level of burden of treatment deprives families of personal (e.g. engaging in leisure activities) and social (e.g. sustaining friendships) resources that cannot be used for the challenges at hand (including participating in SDM), which, eventually, amplifies a child's vulnerability. Another essential finding is that leukemia patients are confronted with an even higher burden of treatment, as measured by number of inpatient stays, inpatient visit duration, and the likelihood of dying in the hospital (chapter 4). This translates itself into a higher burden for the entire family. It is important not to (unintentionally) underestimate leukemia patients' burden of treatment because of their comparably good prognosis[8]. In sum, better understanding a family's burden due to treatment adherence improves the understanding of a family's situation which improves SDM and helps to live up to the bioethical ideal of minimally disruptive medicine[9].

Moreover, two studies report that PPC consultations occurred late in the illness trajectory and palliative care began close to death (chapter 3 and 6). This finding is not in line with recommendations of international health organizations, which recommend that palliative care begins at diagnosis[10-12]. Although PPC initiation should be tailored to each individual child, evidence of late PPC initiation highlights need for improvement at the aggregated population level. To better meet PPC guidelines, barriers to timely initiation need to be overcome. The broad evidence of late PPC initiation suggests that strict adherence to the standards set out by the guidelines may not always correspond to the best interests of the patient at an individual level. Finally, the initiation of PPC occurred even later in the illness trajectory for leukemia patients (chapter 3). While the better prognosis might be the reason for this, practitioners need to be cautious not to withhold the benefits of PPC from this group.

Third, barriers to proper implementation of PPC, but also specific barriers to adequate SDM on PPC were identified. Results revealed the following barriers to SDM on PPC (chapter 7): operational factors (e.g. physicians' lack of time and education) and familial factors (e.g. fear of abandonment, cultural values, misunderstood treatment goals). Lastly, another study employing focus groups with health care professionals (chapter 5) revealed parental misunderstanding of PPC and different cultural backgrounds of families as main barriers to timely implementation of PPC. It is self-evident that the aforementioned barriers are likely to negatively impact decision-making. Once they are known, they need to be tackled and, if possible, overcome. Whereas tackling structural and financial barriers at an institutional level is a costly endeavor, most personal barriers on the part of the involved parties can be overcome with a comparatively small amount of resources. For example, proactively reflecting on (individually and within the team) and subsequently addressing these barriers early in SDM (when talking to the family) could facilitate overcoming them.

1.3 Process of Shared Decision-Making

Reported results show that parents' actual roles in SDM did not match their preferred roles in one-third of the cases (chapter 2), even though physicians were able to correctly assess the preferred roles. Parents' actual role was typically more passive than they wanted. Enabling parents to hold their desired role is crucial because holding less active roles was shown to be associated with lower satisfaction with SDM[13]. However, this requires constant assessment of parental roles, both preferred and actual. Related to this, findings on information provision by physicians (chapter 2) offer a consistent pattern of parents rating the amount of information they received as less satisfactory than physicians rated them. Again, this supports the need for practitioners to actively assess whether the proffered information was correctly understood. Having considered this, it appears that physicians need to engage in a proactive, continuous, and critical practice of ensuring that parents are on the same page.

Children's involvement in SDM is predicted by their age and gender (chapter 2). While it is in line with international guidelines that older children are more likely to be involved, the greater involvement of girls, as seen in evidence, is not. The earlier maturation of girls is a popular truism that appears to be backed up by neurobiological evidence that points to faster brain maturation for girls[14]. In some countries, this evidence translates into legislation in which male and female minors are generally considered competent starting at different ages[15]. Subject to further research, the effect of sex on brain maturation underpins the consideration of this variable both at a conceptual as well as at the practical assessment level. Moreover, if additional evidence substantiates faster maturation of female brains, it calls for further debate as to whether this factor should be incorporated in policies and legislation.

In the Swiss medical setting, minor patients from 12 years onwards are mostly expected to be capable of judgement. Still, a considerable minority of children 12 years of age or older were not involved in SDM on treatment-related issues or on PPC (chapter 2 and 3). Despite

recommendations to decide about a child's involvement on a case by case basis, these results suggest a stronger involvement of children who are at least twelve years of age as well as more caution not to underestimate their level of understanding[16]. Finally, one reported study finds that leukemia patients were less likely to receive palliative care (chapter 3). This also means that those patients are deprived of the opportunity to participate in SDM and to exercise their right to be heard. There might be prognostic reasons for not initiating PPC, but these children unfortunately died without the opportunity to benefit from palliative care.

Reported findings on the process of decision-making reveal a spectrum spanning from no involvement of children at all to full participation in SDM (chapter 3). Overall, the diverse pattern – eight types of how decisions are made were identified – indicates that there is no standardized way of SDM and can be interpreted as a hint to fulfilling the decision-making criteria of adaptivity and family-centeredness. However, SDM was the most often utilized approach. This might be a consequence of the imperative for SDM in pediatrics. Conversely, in some cases, information was withheld from children because of explicit parental wishes. Only considering parents' wishes with the implicit assumption of their decisional authority or with the aim to preserve the stability of family structure does not sufficiently appreciate a child's right (and need) to be involved. Generally, in those scenarios, a deliberate weighing of a child's right to be involved and respecting parental preferences is required. However, in cases of children who are deemed capable both respecting parental wishes and prioritizing the preservation of the stability of family structure are hardly justifiable. Denying them the opportunity to participate in SDM impairs their capacity and degrades their trust. Moreover, since making decisions regarding health, illness, and well-being is an inalienable right and a need of a child[17-19], in cases of capable children the principle of involving them should trump the other two principles of respecting parents' wishes and of preserving the stability of family structure and thus, the capable child should be involved.

2 Relating the Empirical and the Normative

As outlined in the background of this thesis, there are several ways to integrate the empirical and the normative. In the following section, the selected approach to empirical bioethics, Critical Applied Ethics (CAE), is discussed in context of the reported findings. The significance of the "is" for the "ought" is also presented.

2.1 The Five Stages of Critical Applied Ethics

In the following, it is illustrated how the five stages of CAE are applied within the reported research on SDM in pediatric oncology[20,21]. According to Leget and colleagues, a bioethical analysis of an ethical problem is a perpetual endeavor characterized by ongoing evaluation and re-evaluation[20]. Sharing this conviction, this thesis neither claims to represent an exhaustive bioethical analysis of SDM in pediatric oncology nor pretends to sufficiently explore the subject at all five stages. Rather, it offers valuable contributions to the overall bioethical analysis of SDM in pediatric oncology at various stages (of CAE).

1. Determination of the Problem

Ethically relevant shortcomings surrounding SDM in pediatric oncology were outlined in the first chapter (1.3.1): a dearth of knowledge on ways to realize favorable participation of the child, limited inclusion of children, unfulfillment of preferred parental roles in decision-making, late or non-referrals to PPC, lack of formalized training for physicians. It is important to note that already at this first stage, empirical and normative analyses co-determined the subject of subsequent research. The ethico-legal imperative (1.1.3) provided the normative background against which medical practice in pediatric oncology can be evaluated based on empirical data. Bringing the normative and empirical together resulted in the realization that decision-making in pediatric oncology needs further improvement.

2. Description of the Problem[1]

Once the problem has been identified, it needs to be described in greater detail from both normative and empirical perspectives. The thorough description of (different aspects of) the problem was mainly outlined in the results sections of the included manuscripts. The ethical analysis of (hidden) values and understandings that were imported through concepts and terminology (used to discover the moral problem) dealt with the conceptualizations of decision-making capacity and of PPC. This critical look at the concepts and terminology drew attention to the underlying assumptions that are normatively loaded, for example, an individualistic understanding of autonomy fostering an excessively rationalistic conceptualization of decision-making capacity or the close identification of comprehensive PPC with four core domains of care. In such instances, the concepts used to determine the problem inherently involve normative power.

In addition to the ethical analysis, empirical research at this stage has carefully shown how clinical practice manifests itself. By employing multiple empirical methods, the conducted research was able to "to cross-check, verify or 'fine tune' the applicability of theoretic analyses, guidelines and policies in real world practice"[22,p.206]. Subjects of interest were SDM (while considering pre-existing conditions such as burden of treatment or the parents' and physicians' perspectives with respect to evaluations of children's capacity), and timely integration of PPC; furthermore, based on stakeholders' expertise, the implementation of the rather philosophical definition of PPC was examined. Again, this stage demonstrated the dynamic interaction between normative and empirical contributions.

[1] Important insights into how SDM in Swiss pediatric oncology occurs were already provided by the research that was part of the first phase of the project. However, these results cannot be reported in detail here.

3. Effects and Alternatives

Consequences of decisions or policies were explored within the conducted research. It should be noted that research describing the problem (stage two) and outlining the effects and alternatives (stage three) overlap significantly because describing the current clinical practice and examining the effects of recommendations cannot be fully dissociated. Therefore, both appear primarily in the results and discussion sections of the included manuscripts. Empirical analysis focused on aspects surrounding SDM which are clearly related to an "ought", such as the recommendations to improve child involvement, share decision-making responsibilities (e.g. information provision by physicians, roles of parents), and initiate palliative care in a timely manner. Effects of these recommendations were empirically analyzed, and the respective findings concluded that these standards are not being fully met in clinical practice.

At the same time, normative ethics provided important considerations at this stage. It highlights and counters any premature conclusions drawn from the empirical evidence. For example, in the face of parental wishes to withhold information from their children, one could hastily conclude that given these circumstances, parental authority should be prioritized under all circumstances. Such a dichotomous approach would not accurately capture the full complexity of the ethical issue, and as such, normative ethics prevents from drawing this conclusion: generally, by highlighting the disconnection between moralities that are supported by data and those supported by ethical theory, as well as by referring to concepts like relational autonomy and SDM.

4. Normative Weighing

This stage represents the very nucleus of empirical bioethics. Normative weighing within this thesis took place in the discussion sections of the included manuscripts and is summarized in the first part of this chapter (10.1). Empirical analysis contextualized the respective normative

theories and provided actual experiences from medical practice. For example, by exploring pre-existing conditions of SDM, empirical analysis refers to aspects which need to be taken into account by normative considerations on SDM.

Normative ethics, in turn, warns against an unconditional acceptance of the status quo. In the face of late initiation of or even non-referrals to palliative care, it urges towards changing this practice in order to come closer to the articulated "ought". Further, normative theory challenges statistical normativity as it "stands over and against the idea that the majority [a given clinical practice] creates the moral rules"[20,p.233]. On the other hand, empirical data informs normative theory and determines ethical conclusions through a balancing process that seeks coherence of relevant considerations (e.g. empirical, theoretical) and that meets the criteria of rationality and consistency[23]. In particular, bearing in mind the ethico-legal imperative for SDM, this thesis discussed the empirical findings, and thereby, evaluated the medical practice from this perspective.

5. Evaluation of the Effects of a Decision

This thesis concludes with several recommendations with respect to SDM regarding palliative care in pediatric oncology (10.1.1 - 10.1.3, 10.4). The goal is to improve clinical practice. Research's responsibility is to map out the realized effects of stated recommendations. Such an evaluation of effects needs to be addressed within further research.

Further Remarks

Finally, as recommended by Davies et al., three questions must be discussed within an empirical bioethics project[23]. First, the raised ethical concerns (10.1 and 10.4) were justified through coherence of the following aspects: normative theory (1.1.3), empirical results (results of chapters 2-9), and ethical judgements by the research team (discussions of chapters 2-9, as well as 10.1 and 10.4). By *a priori* assigning the same epistemic status to normative

theory and empirical analysis, CAE allows empirical data to inform and potentially refine normative theory. For example, two literature reviews (on timing of palliative care initiation, chapter 6; and on articles that discuss medical guidelines, chapter 7) showed that the normative theories (e.g. WHO: starting from diagnosis onwards) might not always be appropriate within the context of pediatric oncology. Second, within our consultative approach to empirical bioethics, the interdisciplinary research team represents "the thinker" who arrives at ethical concerns. Participants were consulted to gather relevant data, but were not included in the ethical reflection. Third, although the focus was on pediatric oncology, a variety of findings on SDM can be applied to the pediatric setting in general; for example, the differing parental and physician perceptions or the parental perception that information provided by physicians is insufficient.

2.2 The Contribution of "Is" to "Ought"

Building on the typology of Kon[24], the functions of empirical data within this empirical bioethics research project are briefly summarized. The empirical analysis fulfilled four main functions, as will be described in the present section. Detailed descriptions of the four means of contribution appear in the respective chapters.

First, it provided "lay of the land" research by describing the current practice of SDM regarding palliative care in pediatric oncology (results sections of chapters 2, 3, 5, 6). Second, it compared the status quo regarding SDM and the initiation of PPC with an articulated ideal (results and discussion sections of chapters 2, 3, 5, 6). Third, it examined how practice can be improved (discussion sections of chapters 2-9). Fourth, it recommended amendments of normative theories based on the previous three levels (discussion sections of chapters 2-9, as well as 10.1 and 10.4).

3 Limitations and Implications for Further Research

Studies presented in this thesis were conducted as part of the project "*Attitudes and motives concerning end-of-life decisions: Competency and autonomy of children and adolescents in paediatric oncology*." The following sections discuss methodological limitations and implications for further research.

3.1 Limitations

Limitations of the included studies are discussed in the respective chapters (see chapters 2-9). Moreover, there are limitations that stem from the overall mixed-methods approach and from the empirical bioethics methodology.

First, the presented research consists of empirical (quantitative and qualitative) and theoretical (conceptual) findings whose compatibility is not immediately apparent. However, SDM embraces both theoretical constructs (e.g. decision-making capacity) and empirical patterns (e.g. a child's involvement) and thus, must consider both perspectives. The different findings were organized in three separate sections to avoid confusion.

Second, the scope of application must be carefully considered. The reported qualitative results cannot be generalized because the sample cohort did not represent a random sampling from the target population (Swiss pediatric oncology providers) and was limited in number. Still, they offer a contextualized understanding of an insufficiently researched subject, namely pediatric oncology providers' attitudes towards PPC, and can be transferred to other settings by readers who can evaluate the extent to which the findings apply to other contexts[25]. Further, all reported empirical studies were conducted in Switzerland and thus, primarily capture clinical practice of the participating SPOG centers. Despite natio4nal particularities, the reported findings have limited transferability because: (a) studies from

other countries revealed comparable results[13], (b) of a widespread reliance on protocols childhood curative treatment regimes are similar across different countries, and (c) preliminary quantitative results, for example on the dyad-perspectives on the same child, point towards systematic differences between physicians and parents and, therefore, can be generalized to larger populations.

Third, the perspective of children was not consistently captured directly. Yet, children's perspectives were gathered indirectly through surveying parents and physicians. They were also gathered directly in the first part of the project.

Fourth, the relationship between the empirical and the normative represents a challenging task containing many potential pitfalls. Drawing normative conclusions by balancing certain empirical and normative considerations is inherently limited and might leave out other important considerations. Moreover, the set of normative premises that constitute the normative framework was chosen because it represents the relevant ethico-legal background of SDM in pediatric oncology. However, that should not obscure the fact that other normative premises could have been chosen and that those used within this thesis are culturally determined. However, the research team - being culturally and professionally diverse itself - sought to capture the most relevant and internationally accepted normative premises.

Fifth, the author of this thesis has a background in Psychology and Applied Ethics, which might impact the planning and conducting of the research, as well as the discussion of the results. Yet, this constricted perspective was counterbalanced through contributions from and constant discussions with other members of the research team who had diverse backgrounds and varied epistemological and ontological stances (e.g. Philosophy, Gerontology, Biomedicine, Medicine, Psychology, Theology, Bioethics, and Statistics).

3.2 Implications for Further Research

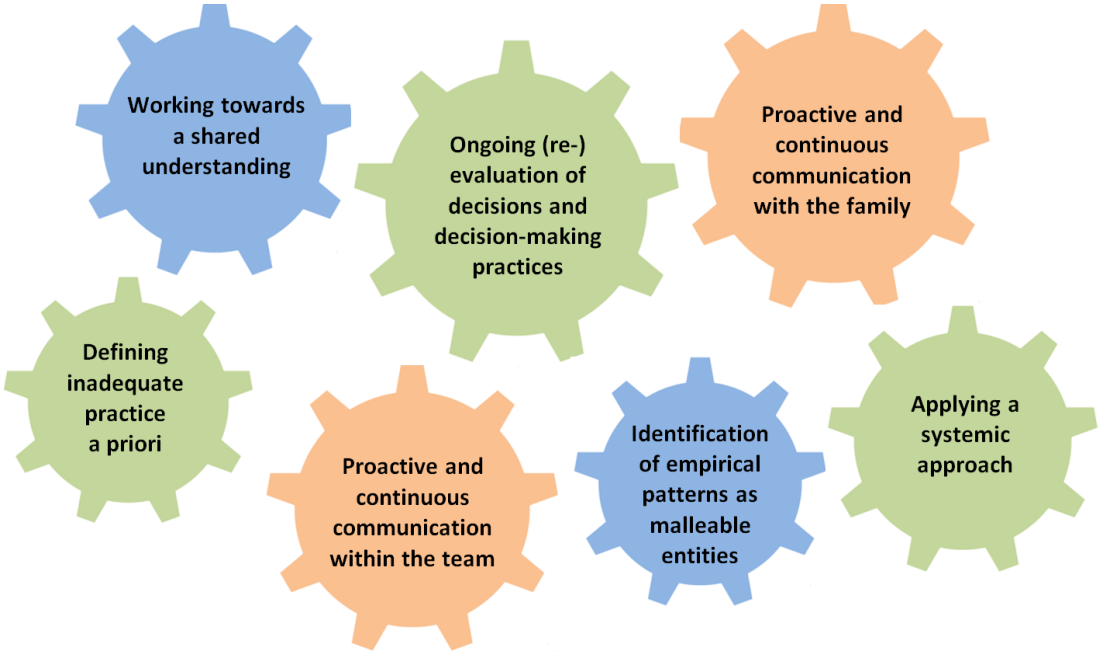
This thesis provides numerous recommendations (10.1.1 - 10.1.3, 10.4) for clinical practice and further research. It is of cardinal importance to map out the effects of the respective recommendations.

At the theoretical level, more research is needed to explore concepts linked to SDM, such as decision-making capacity, and the question of what is understood by the term PPC among different occupational groups and across different countries. Conceptual clarification is necessary to achieve a shared understanding, which facilitates the actual provision of PPC (including SDM) and research on these issues. Agreement on the terms also fosters the acceptance of PPC among the public. Second, with respect to SDM itself, children's perspectives need to be captured more directly by future research. Only by including children's views and attitudes an applicable model of SDM can be developed. Further, children's decision-making capacity requires further empirical investigation in order to involve them in an adequate way. Third, SDM (and related concepts, such as autonomy and capacity) should be studied within different clinical settings. Reported findings showed that, for example, the evaluation of children's abilities depended on the type of decision (e.g. end-of-life versus related to curative treatment). Therefore, approaches to SDM need to be grounded in the respective clinical setting. Finally, future research should develop evidence-based tools for clinical practice to support practitioners in face of their challenging tasks (e.g. capacity assessment tools, a vademecum for communication with the family, short reminders of potential pitfalls within SDM), without losing sight of the notion that every child represents an individual case.

4 Conclusions

The overall goal of this thesis was to propose ethically sound and practically feasible ways to facilitate SDM regarding palliative care in pediatric oncology. The preceding chapters provide insights into important aspects of SDM, such as underlying conceptualizations, pre-existing conditions, and the process of SDM itself. This thesis concludes with the formulation of several recommendations for clinical practice and theory (figure 2) which arise from integrated empirical and normative analyses and express the very essence of the findings. Naturally, they are not fully distinct, but partially overlapping.

Figure 2. Recommendations for practice and theory



1. Working towards a Shared Understanding

A shared understanding and a common language among health professionals are a prerequisite for quality PPC and implementable research outcomes because each goal

requires effective dialogue among researchers and practitioners. Working towards an unambiguous use of terms related to PPC among health professionals represents a necessary cornerstone on which to enhance clinical practice. Otherwise, it remains likely that misunderstandings (e.g. between team members, between family and staff, between inpatient and outpatient setting, between policy makers and pediatric oncology providers, between lay public and professionals) will continue to impede development and provision of PPC. Therefore, any efforts to provide PPC should entail clarification of the used terminology.

2. Proactive and Continuous Communication within the Team

Proactive and continuous communication among the team members is crucial to quality PPC as it serves multiple functions that reach beyond regular treatment-related discussions. First, it prevents misunderstandings based on terminological differences and with respect to care responsibilities. Discussing the various and differing understandings helps to establish a shared vision of PPC and facilitates a team's capability to work together. Second, besides fostering care and communication, frequent team meetings can provide a necessary framework for reciprocal supervision. Being able to address uncertainties and burdening experiences is beneficial for mental hygiene among team members. Third, exchanging narratives of experiences improves team collaboration. Fourth, it prevents from neglecting principles of medical ethics by utilizing collective attention to discover cases of violated rights of a child, for example, not including a capable adolescent in decision-making because of explicit parental wishes. The latter case contravenes a key principle of medical ethics and, therefore, presents an opportunity for retrospective evaluation and future improvement. In sum, regularly held round table meetings that not only discuss medical questions, but also offer space for reciprocal supervision and the exchange of narratives should be implemented.

3. Proactive and Continuous Communication with the Family

A proactive and continuous approach to communication with the family is necessary to adequately address potential sources of confusion among the involved parties. For example, with respect to the underlying perspectivity of evaluations of the child and with respect to information provision by physicians, pediatric oncology providers need to assess whether both parties hold similar views of the child and whether parents correctly understood the information provided. Generally, communication is a process between a sender and a recipient influenced by various variables. Even under normal circumstances, communication often fails. In the face of a child's illness or even imminent death, communication is fraught with uncertainty and fear, with systematically differing perceptions between the treating staff and parents, and with misinterpretation. To better detect differing perceptions and misinterpretations, pediatric oncology providers should carefully examine whether the involved parties are on the same page and should not take for granted that what they intended to say matches what the other party understood. A small and continuously updated vademecum on relevant features of communication with the family could be a helpful tool.

4. Ongoing (Re-)Evaluation of Decisions and Decision-Making Practices

SDM regarding palliative care in pediatric oncology currently does not fully meet the standards recommended by international guidelines: involving children in decision-making processes, realizing preferred parental roles, or timely initiation of PPC consultation. Considering these shortcomings, but also because of the high standards for SDM in pediatric oncology, a monitoring system should be installed at two levels. First, at an institutional level, decision-making practices should be regularly discussed within the team and outcomes should be regularly documented in the medical records. Second, at an aggregated and trans-institutional level, decision-making practices and outcomes should be collected and

supervised, for example by a specialized clinical ethicist or similar expert. At both levels, a standardized form for the recording of discussions not only improves record keeping, but at the same time could expedite the evaluation of SDM. Such a two-level system is of course not limited to the evaluation of decision-making practices to address shortcomings. More importantly, it provides simultaneous evaluation of emerging trends and challenges that may require implementation of new or adaption of existing policies. For example, ethical supervision should be developed with tactful respect for parental attitudes that are contrary to their child's best interest, such as possible preferences for herbal supplements over conventional analgesics among parents.

5. Applying a Systemic Approach

Parents and siblings are strongly influenced by the illness of the child and the accompanying burden of treatment. At the same time, children are highly dependent on their parents and social networks. Having considered these aspects of relationality and dependency that are particularly relevant for children, it is evident that the entire family embodies the unit of care and must be recognized as such also in SDM. Thus, pediatric oncology providers should provide care from a family perspective and should see the preservation of the stability of family structure as an important tenet of PPC.

6. Defining Inadequate Practice a priori

It was shown that there is no universal way to provide PPC that can be applied across different children and their families. This is due to the fact that children and their families have highly individual needs that can span various realms, such as physical, psychological, social, or spiritual needs. Therefore, the concrete form of adequate PPC for a particular child cannot be positively (and exhaustively) defined a priori. However, with respect to SDM regarding PPC inadequate practice can be defined a priori. Such ex negativo approaches have

the advantage of clearly determining what is off-limits while maintaining the necessary degrees of freedom for individually tailored care. They define a threshold below which practice becomes ethically unjustifiable, for example excluding a capable adolescent from SDM based on parental wishes, a physician using a single assessment of capacity towards many types of decisions (that require separate assessments), withholding information from parents or children, or disregarding relational aspects of a child's autonomy by exclusively focusing on rationality. By drawing explicit boundaries, an ex negativo approach can facilitate the enforcement of principles of medical ethics. A list of negatively defined principles about what not to do could serve as a handy tool for practitioners in their daily clinical practice and as a framework of reference, for example, during multidisciplinary round table meetings.

7. Identification of Empirical Patterns as Malleable Entities

Empirical patterns related to SDM in pediatric oncology, such as the decision-making capacity of a child, parental preferences regarding SDM, a child's condition, or a child's participation, are dynamic and constantly changing over time as well as across situations. Consequently, applying this awareness to clinical practice means to scrutinize universal statements based on inductive generalizations and to instead refer to situational statements that relate to the specific circumstances, such as the type of decision in case of capacity assessment. Moreover, dynamic empirical patterns need to be captured by fluid conceptualizations. Otherwise, they remain hardly applicable and misleading. Therefore, practitioners should understand empirical patterns related to SDM as malleable and should be cautious with universal statements regarding the latter.

These closing recommendations reflect the complexity of the presented research subject as well as the difficulty to shape decision-making in an ethically sound way that satisfies all

parties involved. However, beyond any attempt to facilitate the SDM process, there remains a residual of SDM regarding palliative care in pediatric oncology that appears immune to any form of facilitation. When a child is diagnosed with cancer, parents and the child are confronted with death and dying and sometimes go through anticipatory grieving. This situation engenders overwhelming emotions. Moreover, the prognostic uncertainty of childhood cancer creates a brutal vacillation between two realities along the illness trajectory: hope and despair. Today, almost one in five children dies due to cancer. However, children are supposed to outlive their parents, they "are not supposed to die, but they do"[26,p.10]. Losing a child to cancer is an unutterably traumatic experience which touches the deepest and most profound layers of human existence and which parents metaphorically described as "being covered in a wet and dark blanket".

> The blanket was already present and covered the parents when the child was in its palliative phase. To see the child suffer was emotionally arduous, but facing the child's imminent death could be even more difficult. (…) The grief was at this moment [of death] intensified and thus the blanket was experienced to become even wetter and darker. (…) Gradually, the blanket dried little by little, but it still felt overwhelming. Sometimes, when emotionally hard memories came forth, it "rained" grief and pain and the blanket would once again become more wet, dark and heavy. With time, the rain did not come as often as before. A way of handling the grief could be to live in the moment since the future felt too uncertain. As years pass, the blanket became drier and life became brighter and less oppressed, but in some ways it would always be present.[27,p.42]

All this testifies of the existentiality of experiencing childhood cancer. With that in mind, this thesis acknowledges the boundaries of the presented research. Still, as demonstrated throughout this thesis, sharing the process of decision-making is one ethically sound way to reduce burden on the involved parties. Correspondingly, this thesis was written in the modest and compassionate hope of facilitating the sharing of decision-making in face of childhood cancer and, ultimately, of reducing burden for children with cancer and their families.

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Appendix

Interview Guide for Focus Groups (Chapter 5)

Attitudes and motives concerning end-of-life decisions:
Competency and autonomy of children and adolescents in paediatric oncology

A. INTRODUCE US AND THE PROJECT

B. SET THE GROUND RULES:

C. INTRODUCTION

D. FOCUS GROUP QUESTIONS:

1. Since the goal of the project is to understand palliative care in Swiss pediatric oncology, we would like to start by knowing YOUR understanding of pediatric palliative care.

2. Could you please tell us how pediatric palliative care is defined in your SPOG?
    a. Is there a protocol or guideline in place in your SPOG?
    b. Ask about supportive care.
    c. Specific palliative care teams.
    d. Multidisciplinary care
    e. Integrative approach

3. Please describe the most recent patient whom you referred to palliative case?

4. Thinking about the same case, what were according to you the difficulties/barriers to implement PPC?

5. When and under what circumstances do you meet with an ethical committee in order to discuss the further treatment or shift to EOL care (or to PPC care exclusively)?

6. Now, we are interested in knowing the process of communication (with the family: parents and children) that happens:
    a. First, when the information is given to them that the patient is no longer curative and palliative care could be provided.

   b. Second, the decisions that take place in organizing continued care at SPOG, at home and other alternative treatments.

7. What role does the family have in these decisions?
   a. Is this role same as it has always been during the course of earlier treatment?

8. We have prepared a short case for you to read so that we can discuss decision to enter palliative care and what happens thereafter.

9. What do you think should be done to improve palliative care in pediatric oncology?
   a. Mention WHO palliative guideline.

10. Is there anything that we have not discussed, but you would like to add?

Questionnaires (Chapter 2)

1. Parents

Lieber Teilnehmer/in: Vielen Dank für Ihre Zustimmung zur Teilnahme an dieser Studie und ebenfalls dafür, dass Sie dazu bereit sind Informationen über die Erkrankung Ihres Kindes mit uns zu teilen. Die Informationen, die wir durch diese Studie zu erwerben hoffen, sind sehr wertvoll, da sie uns helfen zu verstehen, wie Kinder in der Schweiz mit Krebs leben. Insbesondere die Art und Weise wie Sie ihr Kind in den Entscheidungsprozess zur Behandlung einbezogen haben, wird anderen Eltern sowie betreuenden Ärzten in einer ähnlichen Situation möglicherweise helfen. Bitte vervollständigen Sie die Angaben in diesem Fragebogen zu Ihnen und Ihrem Kind, welches entweder in einem der Schweizer Pädiatrischen Onkologie Zentren diagnostiziert und/oder behandelt wird/wurde. Die Fragen behandeln Daten zu Ihrer Person, zur Diagnose und Prognose Ihres Kindes und zu eventuellen sterbebegleitenden Behandlungen. Selbstverständlich werden Ihre Daten und Antworten anonym und vertraulich behandelt, da wir nur ihren Identifizierungscode benutzen werden und nie Ihren Namen oder andere Informationen.

Instruktion zum Ausfüllen des Fragebogens: Bitte beantworten Sie die folgenden Fragen bezüglich ihres Kindes. Kreuzen Sie die Antworten an, die am besten passen.

Eltern Identifizierungscode:

Grund für den heutigen Besuch:

☐ Mein Kind hat eine Krebsdiagnose erhalten (neuer Fall) ☐ Kontroll- oder Behandlungstermin

1. Demografische Informationen für Eltern:

a. Alter: _____ Geburtsjahr: _____

b. Geschlecht: ☐ männlich ☐ weiblich

c. Nationalität: ☐ Schweizer/in ☐ Andere – Herkunftsland:

d. Sprachen:

e. Familienstand: ☐ verheiratet ☐ geschieden ☐ verwitwet ☐ ledig ☐ anderes (bitte angeben):

f. Religion: ☐ katholisch ☐ evangelisch ☐ andere (bitte angeben):

g. Ausbildung: ☐ keine Ausbildung ☐ Berufsausbildung ☐ Abitur/Matura ☐ Fachhochschulabschluss ☐ Universitätsabschluss

h. Anzahl der Kinder:

j. Bitte beschreiben Sie Ihre Arbeitssituation und die Ihres Ehepartners:
☐ beide vollzeitbeschäftigt ☐ ein Partner vollzeitbeschäftigt, der andere in Teilzeit,
☐ ein Partner vollzeitbeschäftigt und der andere komplett Zuhause
☐ beide Partner in Teilzeit beschäftigt ☐ Selbständig

2. Generelle und krankheitsbezogene Informationen über Ihr Kind:

a. Alter: Geburtsjahr:
b. Geschlecht: ☐ männlich ☐ weiblich
c. Nationalität: ☐ Schweizer/in ☐ Andere – Herkunftsland:
d. Datum der Krebsdiagnose: (mm/jjjj)
e. Art der Diagnose (Krebsart):
f. Derzeitige Prognose: ☐ ausgezeichnet ☐ sehr gut ☐ gut ☐ schlecht ☐ sehr schlecht
g. Wer wurde über die Diagnose informiert?
☐ nur ich ☐ ich und mein Partner ☐ mein Kind und ich ☐ alle zusammen ☐ Dritte
h. Wer wurde über die Prognose informiert?
☐ nur ich ☐ ich und mein Partner ☐ mein Kind und ich ☐ alle zusammen ☐ Dritte
i. Bitte kreuzen Sie an, welche der nachfolgenden Behandlungen empfohlen wurden:
☐ Chemotherapie ☐ Bestrahlung ☐ monoklonale Antikörper
☐ Operation ☐ Knochenmarktransplantation
☐ andere (bitte angeben):
j. Mein Kind ist in Behandlung seit:
☐ <1 Monat ☐ 1 – 6 Monate ☐ 7-12 Monate
☐ 1 – 2 Jahre ☐ 2 – 4 Jahre ☐ Länger als 4 Jahre
k. Die erwartete Behandlungsdauer für mein Kind beträgt:
☐ <1 Jahr ☐ 1 – 2 Jahre ☐ 2 – 4 Jahre
☐ Länger als 4 Jahre ☐ keine Angabe

l. Bitte kreuzen Sie an, welche Symptome mit der Erkrankung Ihres Kindes einhergehen:
☐ Schmerzen ☐ Müdigkeit ☐ Verstopfung
☐ Übelkeit ☐ Durchfall ☐ Schwindel
☐ vermindertes Bewusstsein/Benommenheit ☐ Schwäche
☐ andere Symptome (bitte angeben: neurologisch, das Herz betreffend, Atmung, etc):
m. Bitte bewerten Sie das mit den Symptomen verbundene Leid ihres Kindes
☐ erheblich ☐ stark ☐ etwas ☐ wenig ☐ keins

n. Leidet Ihr Kind an einer weiteren Erkrankung (bitte angeben):
o. Geht Ihr Kind noch zur Schule? (Grundschule, weiterführende Schule etc.) ☐ ja ☐ nein

3. Bitte bewerten Sie die Qualität der Informationen, die Sie von Ihrem Arzt erhalten haben:

Information Type	**Vollständige Information**	**Nahezu vollständige Information**	**Information erfolgte teilweise**	**Sehr begrenzte Information**	**Keine Information**	**Bitte geben Sie an, ob Ihr Kind während diesen Gesprächen zugegen war.**	
Diagnose						☐ ja	☐ nein
Behandlungsoptionen						☐ ja	☐ nein
Prognose						☐ ja	☐ nein
Krebsursache						☐ ja	☐ nein
Ansprache auf die Behandlung						☐ ja	☐ nein
Teilnahme an klinischen Studien*						☐ ja	☐ nein

*Eine klinische Studie dient dem Zwecke der medizinischen Forschung, um die Sicherheit und Wirkung von neuen Medikamenten oder Geräten zu testen.

4. Bitte bewerten Sie die Fähigkeiten Ihres Kindes, die folgenden Punkte zu verstehen:

Mein Kind kann/ist	Stimme absolut zu	Stimme zu	Weder noch	Lehne ab	Ich widerspreche
die erhaltene Diagnose verstehen					
die erhaltene Prognose verstehen					
die Ursache für die Erkrankung verstehen					
die Informationen zur Ansprache auf die Behandlung verstehen					
reif genug, um Entscheidungen bezüglich der eigenen Behandlung zu treffen					
reif genug, um über die Teilnahme an einer klinischen Studien zu entscheiden					

5. Wurde die Erkrankung Ihres Kindes als lebensbedrohlich eingeschätzt (Einschränkung der Lebenszeit):

☐ ja ☐ nein
☐ ich weiss es nicht ☐ ich möchte es nicht wissen

6. Bitte beantworten Sie die folgenden Fragen:

a. Welche Behandlungsentscheidungen wurden bisher getroffen:
☐ Chemotherapie ☐ Bestrahlung ☐ monoklonale Antikörper
☐ Operation ☐ Knochenmarktransplantation
☐ Andere (bitte angeben)

b. Im ersten Schritt kreuzen Sie bitte an, wer bei der Entscheidung anwesend war.
☐ Eltern, Kind und Onkologe ☐ Eltern und Onkologe ☐ Kind und Onkologe
☐ Eltern und Kind ☐ nur die Eltern ☐ nur der Onkologe

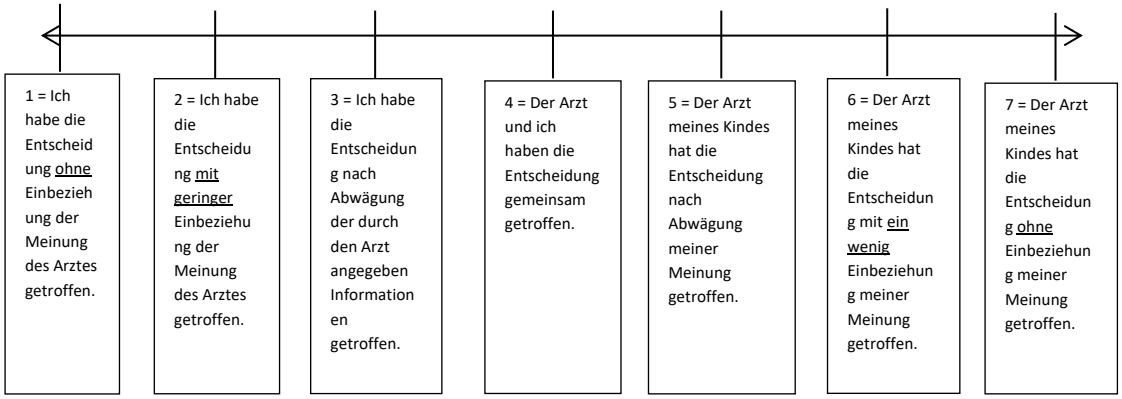
c. Wie zufrieden sind SIE mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

d. Wie zufrieden war der Arzt mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

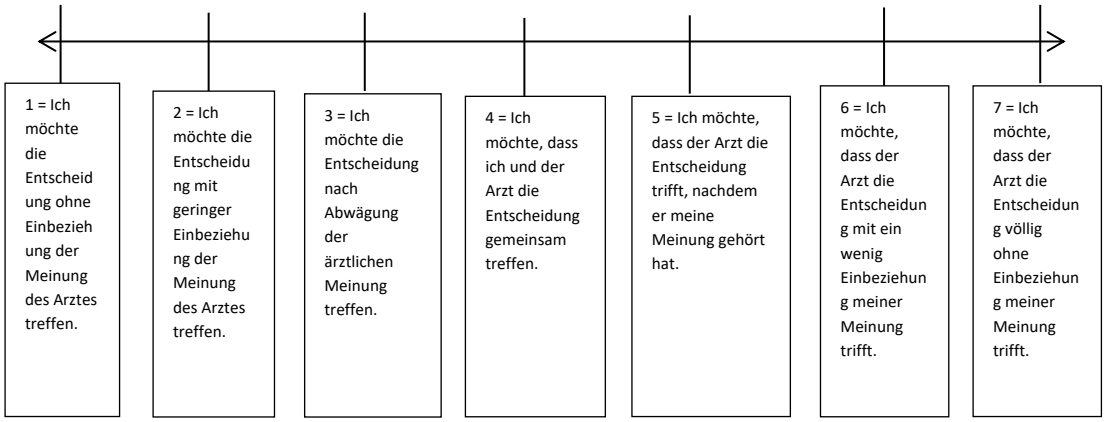
e. Wie zufrieden war Ihr Kind mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

7. Rolle der Eltern im Entscheidungsprozess (bitte angeben welche Entscheidung)

a. Tatsächliche Entscheidungsfindung: Bitte kreisen Sie die Antwort, die auf Sie zutrifft.



b: Bevorzugte Entscheidungsfindung: Bitte kreisen Sie die Antwort ein, die auf Sie zutrifft.



c: Die Entscheidungen sollten gleichberechtigt vom Kind, dem Arzt und den Eltern getroffen werden:

☐ stimme völlig zu ☐ stimme zu ☐ weder noch ☐ lehne ab ☐ lehne völlig ab

2. Physicians

Lieber Teilnehmer/in: Vielen Dank, dass Sie sich entschlossen haben an dieser Studie teilzunehmen und uns mit studienrelevanten Informationen zu versorgen. Die Informationen, die wir durch diese Studie erwerben sind möglicherweise wertvoll, da sie uns helfen zu verstehen, wie Kinder in der Schweiz mit Krebs leben. Bitte nutzen Sie den Fragebogen für alle Ihnen bekannten Fälle von Krebserkrankungen bei Kindern (keine Altersbeschränkung), die entweder in Ihrer Institution in der Zeit von dd/mm/2012 – dd/mm/2013 behandelt und/oder diagnostiziert wurden. Die kommenden Fragen erfassen demografische Daten, die Diagnose des Kindes, die Prognose und Entscheidungen am Lebensende. Selbstverständlich werden Ihre Daten und Antworten anonym sowie vertraulich behandelt, da wir nur ihren Identifizierungscode benutzen werden und nie Ihren Namen oder andere Informationen.

Instruktion zum Ausfüllen des Fragebogens: Bitte beantworten Sie die folgenden Fragen bezüglich ihres Patienten. Kreuzen Sie die Antworten an, die am besten passen.

Arzt Identifizierungscode:

Grund für den heutigen Besuch:

☐ Patient hat Krebsdiagnose erhalten (neuer Fall) ☐ Kontroll- oder Behandlungstermin

1. Patienteninformation

a) Alter: Geburtsjahr:
b) Geschlecht: ☐ männlich ☐ weiblich
c) Nationalität: ☐ Schweizer/in ☐ Andere – Herkunftsland:

2. Informationen bezüglich der Erkrankung des Patienten

p. Datum der Krebsdiagnose: (mm/jjjj)
q. Art der Diagnose (Krebsart):
r. Derzeitige Prognose: ☐ ausgezeichnet ☐ sehr gut ☐ gut ☐ schlecht ☐ sehr schlecht
s. Wer wurde über die Diagnose informiert?
☐ ein Elternteil ☐ beide Eltern ☐ ein Elternteil und das Kind ☐ beide Eltern und das Kind ☐ Dritte
t. Wer wurde über die Prognose informiert?
☐ ein Elternteil ☐ beide Eltern ☐ ein Elternteil und das Kind ☐ beide Eltern und das Kind ☐ Dritte
u. Bitte kreuzen Sie an, welche der nachfolgenden Behandlungen empfohlen wurden:
☐ Chemotherapie ☐ Bestrahlung ☐ monoklonale Antikörper
☐ Operation ☐ Knochenmarktransplantation
☐ andere (bitte angeben):
v. Der Patient ist in Behandlung seit:
☐ <1 Monat ☐ 1 – 6 Monate ☐ 7-12 Monate
☐ 1 – 2 Jahre ☐ 2 – 4 Jahre ☐ Länger als 4 Jahre
w. Die erwartete Behandlungsdauer für den Patienten beträgt:
☐ <1 Jahr ☐ 1 – 2 Jahre ☐ 2 – 4 Jahre

☐ Länger als 4 Jahre ☐ keine Angabe

x. Bitte kreuzen Sie an, welche Symptome mit der Erkrankung des Patienten einhergehen:
☐ Schmerzen ☐ Müdigkeit ☐ Verstopfung
☐ Übelkeit ☐ Durchfall ☐ Schwindel
☐ Bewusstlosigkeit ☐ Schwäche
☐ andere Symptome (bitte angeben: neurologisch, das Herz betreffend, Atmung, etc):

y. Bitte bewerten Sie das mit den Symptomen verbundene Leid des Patienten:
☐ erheblich ☐ stark ☐ etwas ☐ wenig ☐ keins

z. Leidet der Patient an einer weiteren Erkrankung (bitte angeben):

aa. Geht der Patient noch zur Schule? (Grundschule, weiterführende Schule etc.): ☐ ja ☐ nein

bb. Es ☐ arbeiten beide Eltern Vollzeit, ☐ ein Elternteil arbeitet Vollzeit, während der andere Teilzeit beschäftigt ist, ☐ Ein Elternteil arbeitet Vollzeit, der andere Partner bleibt komplett Zuhause
☐ beide Partner in Teilzeit beschäftigt.

3. a. Bitte bewerten Sie den Umfang der Informationen, die Sie den Patienten mitgeteilt haben:

Information Type	**Vollständige Information**	**Nahezu vollständige Information**	**Information erfolgte teilweise**	**Sehr begrenzte Information**	**Keine Information**	**Bitte geben Sie an, ob der Patient während diesen Gesprächen zugegen war.**	
Diagnose						☐ ja	☐ nein
Behandlungsoptionen						☐ ja	☐ nein
Prognose						☐ ja	☐ nein
Krebsursache						☐ ja	☐ nein
Ansprache auf die Behandlung						☐ ja	☐ nein
Teilnahme an klinischen Studien						☐ ja	☐ nein

b. Bitte bewerten Sie die Fähigkeiten des Patienten, die folgenden Punkte zu verstehen:

Der Patient kann/ist:	Stimme absolut zu	Stimme zu	Weder noch	Lehne ab	Ich widerspreche
die erhaltene Diagnose verstehen					
die erhaltene Prognose verstehen					
die Ursache für die Erkrankung verstehen					
die Ansprache auf die Behandlung verstehen					
fähig, um Entscheidungen bezüglich der eigenen Behandlung zu treffen					
fähig, um über die Teilnahme an klinischen Studien zu entscheiden					

4. Wurde die Erkrankung des Patienten als lebensbedrohlich eingeschätzt (Limitierung der Lebenszeit):
☐ ja *(falls ja, überspringen Sie bitte Frage 5)* ☐ nein

5. Falls der Patient eine kurative Behandlung für eine <u>nicht lebensbedrohliche Krebserkrankung</u> erhält, bitte beantworten Sie die folgenden Fragen:

a. Welche Behandlungsentscheidungen wurden bisher getroffen:
☐ Chemotherapie ☐ Bestrahlung ☐ monoklonale Antikörper
☐ Operation ☐ Knochenmarktransplantation
☐ Andere (bitte angeben)

b. Kreuzen Sie bitte an, wer bei der Entscheidung anwesend war.
☐ Eltern, Patient und Onkologe ☐ Eltern und Onkologe ☐ Patient und Onkologe
☐ Eltern und Patient ☐ nur die Eltern ☐ nur der Onkologe

c. Wie zufrieden sind <u>SIE</u> mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

d. Wie zufrieden waren <u>die Eltern</u> mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

e. Wie zufrieden war <u>der Patient</u> mit der getroffenen Entscheidung (1=sehr zufrieden; 5=sehr unzufrieden):

6. Bitte beantworten Sie die folgenden Fragen, falls der Patient <u>sterbebegleitende Behandlungen oder Palliativpflege erhält:</u>

a. Gibt es andere Behandlungsmöglichkeiten: ☐ ja ☐ nein

b. Falls ja, welche:

c. Wäre eine andere Behandlungsmöglichkeit hilfreich: ☐ ja ☐ nein

d. Nimmt/nahm der Patient an einer klinischen Studie teil: ☐ ja ☐ nein

e. Falls ja, welche:

f. Sind die Vorteile von klinischen Studien bekannt: <u>Eltern</u>: ☐ ja ☐ nein; <u>Patient</u>: ☐ ja ☐ nein

g. Haben Sie sterbebegleitende Themen diskutiert, mit: <u>Eltern</u>: ☐ ja ☐ nein; <u>Patient</u>: ☐ ja ☐ nein

h. Falls eine der folgenden Entscheidungen getroffen wurde, kreuzen Sie bitte an von wem und mit wem. Bitte bewerten Sie die Möglichkeiten zusätzlich.

Art der Entscheidung	Ich	Patient	Eltern	Bewertung 1 bis 7. 1= sehr wichtig; 7= unwichtig	Zufriedenheit mit der getroffenen Entscheidung: 1=sehr zufrieden; 5=sehr unzufrieden
1. Stopp der Medikamentengabe					
2. Entscheidung zur Hospitzpflege					
3. Entscheidung zur Palliativpflege					
4. Teilnahme an klinischen Studien					
5. Stopp von lebenserhaltenden Massnahmen					
6. Krebsbehandlungsart					
7. Verzicht auf Reanimation (DNR)					
8. Andere, getroffene Entscheidungen (bitte angeben)					

i. Wurden Massnahmen zur Vorbereitung des Lebensendes angeboten, akzeptiert oder abgelehnt?

Service:	angeboten	akzeptiert	abgelehnt	Gründe für die Ablehnung
1. Palliativpflege				
2. Hospitzpflege				
3. Pflege Zuhause				
4. Intensivpflege				
5. Psychologische Betreuung				
6. Trauerbewältigung				
7. Andere (bitte angeben)				

7. Ihre demografischen Daten:

i. Alter: Geburtsjahr:

j. Geschlecht: ☐ männlich ☐ weiblich

k. Nationalität: ☐ Schweizer/in ☐ Andere – Herkunftsland:

l. Sprachen: ☐ deutsch ☐ französisch ☐ italienisch ☐ englisch ☐ andere (bitte angeben):

m. Familienstand: ☐ verheiratet ☐ geschieden ☐ verwitwet

n. ☐ unverheiratet ☐ anderes (bitte angeben):

o. Bitte geben Sie Ihr medizinisches Spezialgebiet an:

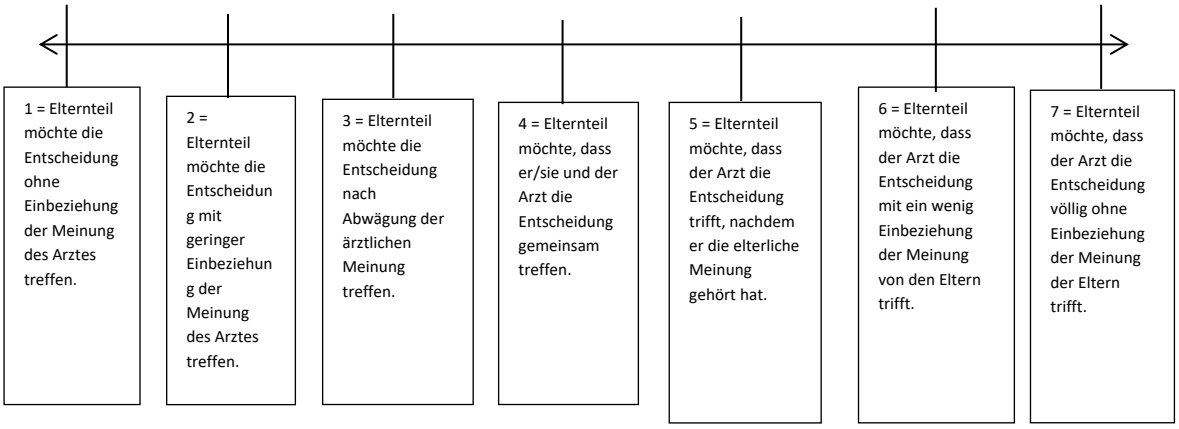
p. Wie viele Jahre haben Sie Erfahrung im Bereich pädiatrische Onkologie:

☐ 0 – 4 Jahre ☐ 5 – 8 Jahre ☐ 9 – 12 Jahre ☐ Mehr als 12 Jahre

q. Religion: ☐ katholisch ☐ evangelisch ☐ andere (bitte angeben:)

8. Ihre Einschätzung, welche Rolle Eltern gerne im Entscheidungsprozess bezüglich der weiteren Behandlung Ihres Kindes hätten:

a. Bitte kreisen Sie die Antwort, die Sie für die zutreffendste halten ein.



b. Die Entscheidungen sollten gleichberechtigt vom Patienten, dem Arzt und den Eltern getroffen werden:

☐ stimme völlig zu ☐ stimme zu ☐ weder noch ☐ lehne ab ☐ lehne völlig ab

Medical Records Data Extraction Sheet (Chapter 3, 4)

Medical Records Data Extraction Sheet: End-of-life project Pediatric Oncology

Instruction: Please use this data extraction sheet to record information as indicated in the medical records of each patient.

i. Health and Demographic information

Sex: _____Male _____Female

Month and year of birth:

Initial Diagnosis:

Date (month and year) of diagnosis:

Indicate the tumour stage at diagnosis (include indications of the malignity grade and whether the tumour is localized or metastatic):

Information on Relapse(s)

Total number of relapses:

Specify relapse 1:

Indicate the date (month and year) of 1st relapse(s):

Indicate the tumour stage at 1st relapse(s):

Specify relapse 2:

Indicate the date (month and year) of 2nd relapse:

Indicate the tumour stage at 2nd relapse:

Specify relapse 3:

Indicate the date (month and year) of 3rd relapse:

Indicate the tumour stage at 3rd relapse:

Add information for further relapses:

ii. Treatment Information – list of diagnosis[1]

A1. Indicate whether a protocol was followed at diagnosis:

If yes, indicate which one (name):

If not, which treatment was followed:

Record data what was done as part of that treatment (e.g., cycles of chemotherapy, fractions of radiotherapy, surgery etc.).

Record respective treatment responses (complete, partial, no response, progressive):

Record the side effect(s) of the respective treatments:

A2. Which treatment was followed at 1st relapse:

Indicate treatment name:

Was it an oncological and/or supportive treatment?

Record what was done as part of that treatment (e.g., if supportive: cells, platelets, fluids, pain, and antibiotics provided).

Record respective treatment responses (complete, partial, no response, progressive):

Record the side effect(s) of the respective treatments:

A3. Which treatment was followed at 2nd relapse:

Indicate treatment name:

Was it an oncological treatment and/or supportive treatment?

Record what was done as part of that treatment:

[1] Repeat treatment information based on the number of relapse.

Record respective treatment responses (complete, partial, no response, progressive):

Record the side effect(s) of the respective treatment:

B. Describe the **team discussion** and decisions regarding **treatment**

- at diagnosis:
- at relapse(s):

C. Were other **centres** (both national and international), specialists or tumor boards consulted?:

D. Specify whether the patient followed any **complementary or alternative treatment(s)**:

iii. <u>Transition from curative to palliative care</u>

A. **Is there any clear indication that the disease is progressive**? _____Yes _____No

If yes, please specify:

B. Is there any indication in the course of the treatment of the following**:**

(1) **decreased** intensity of oncological treatment? ____Yes ____No

If yes, please specify:

(2) **increased** intensity of supportive treatment? ____Yes ____No

If yes, please specify:

(3) an **explicit experimental** treatment? ____Yes ____No

If yes, please specify:

(4) **palliative** treatment? ____Yes ____No

If yes, please specify:

(5) **stopping** treatment? ____Yes ____No

If yes, please specify:

C. Was there a consultation with the **pain team?**

If yes, please specify which pain treatment(s) was provided (e.g., pharmacological and non-pharmacological, length of pain treatment):

D. Was there **an AD HOC ethical committee** consultation**?**

If yes, please indicate the date:

and specify:

(1) who was part of the committee:

(2) if there was a Do Not Attempt-Resuscitation order:

(3) if a withdrawal of life-support occurred:

E. Please note any information about the **transition from curative to palliative phase** (determined by physiological factors, parent agreement etc.):

F. Please indicate whether the transition was discussed among the **medical team members:**

G. Was there an indication of discussions with the child's **paediatrician and Spitex?**

If yes, specify:

H. Discussion regarding the **switch to palliative care took place with patient and family**?

_____ Yes (with both patients and parents)

_____ Yes (only with the parents)

_____ No

_____ Data not available

If yes, record all information related to what was discussed:

I. Was there any suggestion (on the part of the physician) or wish (on the part of the child or parent) to go **home**?

J. If **home care was sought,** please indicate information about the **homecare team**:

K. When did death occur and where (month and year)**:**

iv. Further Notes: