Ethical challenges and opportunities in communicating about cancer. Experiences and outcomes of information sharing between physicians, parents and pediatric patients

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Dean

'Toutes les grandes personnes ont d'abord été des enfants.

(Mais peu d'entre elles s'en souviennent.)'

(Antoine de Saint-Exupéry**,** Le petit prince)[1]

[1]Antoine de Saint-Exupéry. Le petit prince. Évreux (Eure), France; Folio, Kapp Graphic; 2015.

For Livia, with love and gratitude

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Summary

Treatment improvements for childhood cancer have resulted in high survival but the ardor of therapy and care experiences, physically and emotionally affects parents and patients. Relationships with clinicians play an important role for supporting them in achieving improved outcomes. Communication is at the center of building trust and also ensuring understanding of information for permission for treatment, both an ethical and legal requirement. However, processes are many times reduced to exchanges rather than sharing of information and there are shortcomings in physician training in efficient and empathic communication. This work discusses communication in medical encounters during cancer treatment and explores the importance of microethics, the everyday actions and interactions, for improving understanding and information sharing. A model of exploration of illness and experiences is proposed for daily use in the clinic to remedy difficulties in diagnosis disclosure and decision-making. This model can facilitate later communication for poor prognosis and discussions on research participation and end-of-life care.

The doctoral thesis starts with a general introduction on issues in cancer care, normative aspects of decision-making in pediatrics, ethical suggestions for patient participation and difficulties of implementation in clinical practice. Subsequent chapters draw from two studies conducted in Swiss and Romanian pediatric oncology units to address issues related to parental and physician experiences during communication and in involving children and adolescents in their care.

Chapter I offers different perspectives on communication in pediatric oncology and identifies diagnosis disclosure and the involvement of patients as particularly challenging. Attention is paid to analyze physician practices in relation to ethical principles and to identify professional duties.

Chapter II explores decision-making processes and the inclusion of patients in care in Switzerland and Romania. It starts with a theoretical analysis of legal and ethical aspects of capacity and then moves to provide empirical data on how parents decide for their children and what contribution they, their children and physician can make in the process.

Chapter III is a theoretical exploration of ethical justifications for the involvement of minor patients in research that does not aim to benefit the individual patient, but to provide generalizable knowledge. The first article of the chapter argues that patients can have group interests and therefore contribute to promoting them without seeking personal benefit. Then, a phenomenological theory of children as body-subjects further highlights ethical reasons for allowing non-beneficial research in pediatrics. A study exploring minor patients' views on cancer care adds to the argument that research with children is necessary for improving health care outcomes.

A last Chapter summarizes a literature review on end-of-life decision-making and discusses sensitive issues in communicating with patients at the end-of-life by affirming the importance of including children's voices.

The general discussion brings all empirical and theoretical findings together to analyze communication in pediatric cancer care and proposes a model of exploration (EMICPO) to improve outcomes for parents and patients. Further implications for clinical practice and future research are presented.

Although the results of this work focus on difficulties and failures to appropriately communicate in cancer care, its broader goal is to offer reassurance that with the help of physicians and through increased awareness of parental and child experiences, families and patients can get through cancer.

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Introduction

Few illnesses evoke for patients and their families so many fears and different scenarios dominated by uncertainty as a cancer diagnosis[1-3]. Normal daily-life routines are disrupted for the whole tribe and the consequences of living through the cancer experience are long felt[3-8]. Different portrayals of cancer, its treatment and what meaning they have for patients and their families permeate social discourses. Rebecca Dresser talks about a "public picture of cancer [that] is incomplete"[1], usually oscillating between openness about one's own cancer journey in the public sphere and avoidance. Similarly, media representations and therefore society's views of cancer are many times shaped around the "battling" and "fighting" images which convey that rather than being patients, people suffering from cancer are or will eventually become heroes, survivors[1,2,9]. These depictions may stand in stark contrast with feelings of being overwhelmed, the isolation and anguish patients and their loved ones experience[3,10-13]. Even more, cancer for many equates with a less hopeful image - that of a dying patient - and symptoms or even suspicion of cancer trigger heightened anxiety and existential questioning[1,14,15].

Childhood cancer is particularly difficult, as the diagnosis comes at a time when life is just beginning. It challenges parents to confront an existential crisis defined not only by their own fragility, but also their child's and poses a threat to their parenting[2,14,16,17]. Physicians caring for children suffering from cancer have a unique position as they can offer patients and their parents information about the diagnosis and treatment and help them navigate what is happening. As such, clinicians themselves become part of families' intimate journeys through cancer and time and time again have to handle the delicate ways of communicating distressing news and sustaining hope[18-22].

The good, the bad and the ugly in childhood cancer

Prognosis for children with cancer has continuously improved in the past decades and currently childhood cancer survival is at approximately 80%[23,24]. A majority of those diagnosed today will survive as a result of significant advances in research in adults and children[25,26]. The pediatric oncology practice has always been characterized by a strong interweave of standard care and clinical trials protocols[27]. Medical recommendation of treatment is focused primarily on the likelihood of cancer-directed therapy to cure children and improve survival[28]. However, with continuous efficacy improvements treatments protocols have also become more intense, combining various forms of therapies: chemotherapy, surgery and/or radiotherapy. Cancer treatment regimens add to illness' burden inducing a large spectrum of physical and psycho-social symptoms - mucositis, dyspnea, drowsiness, lack of appetite, depression, anxiety and fear[29,30]. Therapy itself induces a great deal of pain and many patients report this to be worse than cancer related pain, thus causing significant distress[31-33]. Even after remission is achieved, those children and teenagers who enter the survivorship phase find themselves at risk of developing short and long-term complications or other illnesses, including secondary tumors, as a late treatment adverse-effect[34-37]. All the complications experienced either during treatment or in remission affect the quality of life of children and adolescents.

Parents share many of the pediatric patients' burdens. They are usually the first informed about the cancer diagnosis[38,39]. This communication has a significant impact on them and their reaction is described in several studies as a "blow" that leaves them "breaking down"[38,40]. After receiving the diagnosis and due to their main role as caregivers accompanying children, parents endure significant stress and emotional burdens. This can take a toll on their psycho-social wellbeing and expose parents to a series of posttraumatic symptoms which may linger in the post-treatment years[41,42]. The presence of symptoms in

parents at diagnosis time raises questions regarding parents' wellbeing, adequate ways of communicating distressing information to them and the circumstances parents face when making complex decisions for their children[41,43]. This can directly influence their abilities to involve children and emphasize their own dependency on physicians, but most importantly highlights struggles to support patients' participation in care[38,40,44].

Childhood cancer is therefore a stressful and sometimes traumatic experience for both patients and their families. It puts patients and families under considerable pressure: invasive procedures, long hospitalization periods, various treatment side-effects, loss of control and social disturbances[45-47]. On this background, parents, physicians and patients have to make various decisions regarding children suffering from cancer and their care.

Decision-making in pediatrics

Decision-making processes in pediatric oncology are complex due to the general uncertainty that accompanies cancer treatment and outcomes, but also as a result of choices made available by combining research and care[48-51]. Traditionally, decision-making regarding healthcare for minor patients rests on the assumption of children's incompetence, therefore their lack of mental capacities and abilities to reason about choices, especially those of a complex nature[52,53]. Based on this presumption, the responsibility of making decisions for children lies with physicians, who provide information and medical recommendations, and with parents, charged with giving permission for treatment and care[54,55]. In fulfilling their responsibilities, parents and physicians have to take into consideration two ethical standards: best interest and harm. The principle of best interest of the child is the prevailing standard in making decisions about the course of treatment and is used both by parents and clinicians in promoting patient's welfare[28,56,57].

Decision-making processes are also influenced by the triadic relationships and communication between parents, the medical team and the patient[45,58-60]. In acknowledgment of the child's position in the triad and as a consequence of the evolving recognition of persons' right to self-governance[54,61], the role of minor patients in healthcare has been given more weigh[28,62,63]. As a result, decision-making in pediatrics is increasingly complex and the triad's actors may contribute in different and intertwined ways to the process of deciding for oncological treatment and care[59,60,64].

Physician role in decision-making

Collaboration with patients and parents is fundamental to making decisions[28]. Physicians' primary role is that of a medical expert, who can establish a diagnosis, propose a course of action, and offer information on the illness and possible choices of treatment[59,60]. In this process, individual treatment and care recommendations are subjected to physicians' professional and personal judgment and, in some cases, decisions by the physician are warranted prior to discussions with families[28,56,57,63]. Oncologists can make use of therapeutic privilege and select a course of therapy to propose to parents and families without presenting all treatment options[60,65]. Levine and colleagues[66] argue that this action is justified when physicians' own assessment promotes best interest and lowers risk of harm to the patient[66]. For example, in the case of young children with brain tumors responsible treatment would be to abstain from the option that may increase chances of survival but leave the patient with significant brain damage and opt for another option that has significant, although lower, survival chances, but avoids undue risk of excessive brain damage to patients[66].

Oncologists' secondary role in decision-making is to support parents and children in their participation in healthcare. This duty is fulfilled by providing information in a manner that can be understood by parents and tailoring communication to patients' abilities, according to mental and emotional maturity, and age. Physicians must also assess parents' and patients'

understanding and willingness to participate in the decision-making process[28,56]. In these assessments, physicians should also ensure that parents can engage in voluntary decision-making, free from coercion or extreme influences that may be external (i.e. pressure from family) or internal (i.e. psycho-social)[28,67]. These elements of information, comprehensiveness and voluntariness are fundamental to obtaining informed consent[28,67].

Parental role in decision-making

Parents are generally viewed as possessing high interests in promoting their children's best interests due to the emotional ties within a family, the common goals and mutual commitments of pursuing the wellbeing of all within the group and adults' self-assumed responsibility to care for minors[68]. Including in healthcare, parents' role is crucial in caring for and protecting their offspring's rights[28]. In most legislations patients who are minors, therefore do not fulfill the minimum age of maturity, usually 18 years old, are generally presumed incompetent to make informed decisions in healthcare[56,69]. However, they still preserve the same rights regarding treatment like all other patients, including the right to be informed and to consent, which are exercised for them by parents or legal representatives who providing permission for care[28,69].

Child's best interest is not the sole foundation for the decision-making process, as children are part of a family in which interests are tightly linked[68]. When parents are in a position to make a decision for which the best interest of the ill child is not obvious, they may consider other competing interests, like the wellbeing of the family as a whole[56]. Ross[68] argues for this constrained parental autonomy that is intrinsic to respecting intimate families' rights, which should not be overruled easily[54,68]. In liberal societies, parents are usually given wide latitude in relation to their values and child-rearing choices and as long as their actions are not neglectful or abusive the same leeway should apply for healthcare decisions[70].

Parental decisional rights are respected pending that they do not conflict with the harm principle and, consequently, the decisions parents make do not lead to serious injury, risk of death, disability or pain for their child[28,68,71]. When the contrary situation occurs and after careful consideration of potential risks for all parties, physicians can take legal measures to overrule the need for parental authorization of treatment[28]. This is particularly the case when parents' religious or spiritual beliefs are the justification behind decisions to reject treatment and therefore endanger the child's life or bodily integrity[71]. Physicians' role in these situations increases and they become advocates for children and their best interests, while protecting patients from considerable harms. This position also comes in consideration of children's developing sense of self and of the fact that they may not share parents' values or are incapable of forming and commit to such strong beliefs on their own[71].

Children's and adolescents' participation in care

Children's involvement in care, including participation in decision-making, can play a significant role in ensuring the delivery of best possible care. This step is attuned to increased recognition of respect for children's developmental autonomy[28,62,72,73], as well as growing evidence of promoting minor patients' wellbeing by supporting them in building resilient mechanisms through participation in care management and decisions[74-76]. As such, various medical associations require physicians to pay attention to and respect individual needs when informing families and children about the cancer diagnosis[28,39,62].

Professional recommendations regarding initial communication with children state that diagnosis disclosure should take place without extensive delays, while trying to honor parental wishes on how to inform children. Special consideration for each patient's mental and psychological state equally should determine how the communication of diagnosis discussion is carried out and the extent of the information provided[39,62]. Parental and

physician agreement and mutual understanding in fulfilling this task is essential for assuring patients. It also is meaningful for building trusting relationships between clinicians and families[39,62]. Providing information regarding the nature of the illness and therapy to children is indicated in view of the benefit of reducing their anxiety, but also with the purpose of obtaining assent and avoiding forcing treatment upon children[28]. The American Academy of Pediatrics[28] similarly emphasizes the relationship that physicians need to build with patients in order to support their participation to the greatest extent that their wishes and capacities dictate. Involving and coaching children and adolescents to participate in their care from the diagnosis onward, might result in increased abilities to deal with later and more serious decisions[77]. Particularly for patients for whom treatment fails, involvement is recommended when addressing care goals and choosing a caring plan no longer aiming at curing the patient[28].

Normative aspects of participation

Legislations in different countries contain diverse prescriptions regarding underage persons' rights to decide for themselves. North American countries and many European states regulate decisions for persons who have not attained the age of maturity, usually set at 18 years, by giving parents authority over child and adolescent care[68,78,79]. There are several exceptions to these provisions based on the mature minor doctrine, recognized the United States (US) and the United Kingdom (UK), and also in consideration of minors' best interests when it comes to accessing care for mental or reproductive health[28,55,78]. Seeking patient assent is permissible under such legal provisions, but minor patients' rights are strictly limited. Refusal to provide assent or the expression of dissent is not binding for medical care that would be significant to patient welfare, but is valid in the context of research participation[28].

Several European countries, such as the Netherlands and the UK, have more permissive legislations concerning minors who demonstrate sufficient mental capacities to make

reasonable decisions[80]. In these countries the approach is to award authorization for treatment to parents and recognize different degrees of capacity in children. As a result, laws formally acknowledge the power of assent and the transition to a more active consent by adolescents, giving them rights to decide, together with their parents or by themselves[78,81-83]. Age limits and a mandate for physician assessment of decision-making capacities are usually the standards for granting children and adolescents the right to consent[81]. Higher age limits alone can sometimes be used to declare a presumption of adult-competency for older adolescents[82]. However, even in the case of adolescents with capacity, their decisions can be overruled if treatment is considered to be in their best interest[78,83]. Therefore, different legal provisions shape the way in which physicians interpret their duties to inform patients and their parents.

Participation and communication in practice

Despite significant changes in acknowledging minor patients' claim to be involved in their own care[28,73], participation in practice is difficult to achieve[45,49,84-86]. First, as described above, involvement of children is subjected to legal provisions regarding decision-making powers and authority in healthcare, which in many countries is assigned to parents[56,81]. Second, children and adolescents may sometimes be deemed to have clinical decision-making capacity for certain treatment or procedures, but not for others[55,78]. Such assessments should be performed by physicians, but procedures to establish whether children are capable of making medical choices are not standardized[54,81]. There is not enough evidence on how to meaningfully determine which children or adolescents have capacity and for what decisions and judgments are more likely to be influenced by clinicians' intuition, be guided by legal standards of maturity and constrained by parental attitudes[81,82]. Communication between the parties therefore plays a major role in assessing minors' abilities and also supporting them to participate in care decisions[54,81,82].

Challenges in physician, parent and patient communication

Ambiguity of ethical and legal imperatives on parental rights and authority to decide for children influences physician-patient open communication[82,87]. Despite guidelines encouraging clinicians to fully communicate with minor patients, there is insufficient guidance on how to achieve this while respecting their wishes and those of their parents[39,65,88]. Patients have individualized preferences and some require direct and truthful provision of information, while others seek selective knowledge[40,45]. Additionally, patient involvement is subjected to parental and physician influences which can be both restrictive and supportive. Several studies report the impact that parental fears have on shielding children from information[44,45,89,90]. Oncologists may perceive parents' reaction and efforts to limit information as impeding them to freely communicate with children, but sometimes adopt similar attitudes towards parents when discussing prognosis[20,91]. Physician training, both formal and through observation of senior clinicians, is often limited and inadequate regarding communication and emotional management upon delivering difficult news[92,93]. Therefore, it is particularly challenging for physicians to individually assess for each patient how much to tell and when[40,45].

While it is believed that open communication with children and families is morally appropriate, evidence on benefits and harms of complete truthfulness with patients and on how it affects parents is limited[94-96]. Particularly at the first discussions about their child's cancer diagnosis parents struggle to absorb information and experience fear while trying to define their caring roles and to protect children[12,90]. Open physician-patient communication can be at odds with parental wishes to be informed separately and to have time to navigate their own emotions before having to comfort their children[38,96]. Patient presence in discussions can lead to parents suppressing their own emotions in an attempt not to scare or confuse patients and does not create an opportunity for parents to prepare themselves for how to tell children. It can also act as a barrier to parent inquiries about what they perceive as

inappropriate to discuss with children[40,96]. Parents' need of emotional support and empathic communication vary and can change in time if treatment fails[43,90]. Although, parents recognize that child presence can psychologically benefit patients by showing them respect, they prefer limiting patient participation in communication that alludes to cancer's severity. Discussions about prognosis or risk of dying are distressful for parents and increase their fears of psychologically harming children[40,90,97].

Communication is subjected also to cultural influences that operate on several dimensions: families' representations of cancer and views on death and dying, diversity of values and possible tensions between physicians' and parents' beliefs, perceived roles and hierarchies of physician-parent interactions, stereotyping and judgment of ethnic groups, language barriers[98-100]. The role of cultural beliefs perhaps is most evident and its significance the more important when healthcare professionals commit to deliver care that is patient and family centered[101]. In situations when cure is no longer achievable, providing such care is highly needed by families. Caring when healing is no longer possible is dependent on how clinicians adapt communication to ensure the provision of effective and compassionate end-of-life services grounded in respect and sensitivity towards patient's and families' wishes is[91,102].

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Research gap

Considering the different legal provisions among states and the difficulties physicians face in implementing ethical standards of open communication and patient involvement in cancer care little is known about clinical practices in different countries. It is unclear how both physicians and parents experience their own participation in decision-making and how fears and assumptions influence these processes. Also, there is no consensus on how children's involvement should be achieved and how to balance parents' and children's needs of involvement.

Study aims

The goals of the doctoral study are enclosed into the main aims of the *Attitudes and motives concerning end-of-life decisions: Competency and autonomy of children and adolescents in pediatric oncology*, a prospective 3 years project funded by the Swiss National Science Foundation (SNSF), Switzerland (National Research Program 67 "End of Life", No. 406740_139283/1):

- to explore the ways in which a child or adolecent living with cancer can be included in her care and decision-making processes
- to examine physicians' and parents' associated motives and attitudes behind their actions to include patients or not.

Given this overarching framework, the doctoral work focuses on investigating the challenges that parents and physicians face in communication, as well as in the involvement of and decision-making for children suffering from cancer on the background of different norms, and diverse institutional and cultural contexts. The doctoral thesis is structured in two parts:

(1) one is based on research study's results regarding diagnosis disclosure and practical attitudes towards communication and patient participation, and (2) the other is a theoretical analysis of parent-physician communication with children and of patients' inclusion in particular aspects of care: research participation and end-of-life.

The empirical data will be used to explore ethical concepts of open communication, fears, participation roles in making collaborative decisions for children and adolescents suffering from cancer and their involvement during different stages of treatment. The theoretical analysis will explore different ways in which children can be included in difficult decisions, particularly end-of-life, and what ethical justification can be offered for the participation of children in research without direct benefits.

Within this scope, the following research questions will be addressed:

- What challenges do physicians and parents face when communicating the diagnosis of cancer to patients and how can physicians ethically address them?
- How does parental and physician communication and attitudes shape the involvement of children at different stages of cancer treatment?
- How does participation and communication in decision-making takes place in countries with different norms?
- What benefits and harms should be considered when limiting information provision to patients, particularly regarding diagnosis, prognosis and end-of-life?
- What ethically relevant elements should be considered when assigning decisional priority and authority in decision-making in pediatric oncology?
- Can children's participation in research with low or no direct benefits (i.e. phase 1 clinical trial for incurable cancer) be ethically justified?

Contributions

The doctoral work is based on empirical and theoretical analysis that resulted in 11 publications – six articles drawing from the qualitative data from the Swiss and Romanian studies, one literature review, and three theoretical articles and one commentary on contentious ethical issues in pediatric practice. Results of these publications have been made possible through collaborative efforts with members of the research team of the SNSF study in Switzerland, as well as with partnering physicians involved in enrollment and data collection. Individual contributions for each publication are detailed below, highlighting Domnita Oana Badarau's work as PhD candidate.

(1) **Domnita O Badarau**, Tenzin Wangmo, Katharina M Ruhe, Ingrid Miron, Anca Colita, Monica Dragomir, Jan Schildmann, Bernice S Elger (2015). Parents' challenges and physicians' tasks in disclosing cancer to children. A qualitative interview study and reflections on professional duties in pediatric oncology. Pediatric Blood & Cancer, 62(12):2177-2182

The article was conceptualized by D Oana Badarau in collaboration with Jan Schildmann and Bernice Elger. Together with Jan Schildmann, D Oana Badarau refined the coding of the data that was carried by the first author together with Tenzin Wangmo and Katharina Ruhe. Doctors Colita, Miron and Dragomir facilitated data collection and helped with participant enrollment in the study. D Oana Badarau took the lead in writing the manuscript and coordinating all authors' input. The manuscript was reviewed by Jan Schildmann before being sent for comments and approval of final version by all authors.

(2) **Domnita O Badarau**, Eva De Clercq, Tenzin Wangmo, Monica Dragomir, Ingrid Miron, Thomas Kühne, Bernice S Elger (2016). Cancer care in Romania: challenges

and pitfalls of children's and adolescents' multifaceted involvement. Journal of Medical Ethics, 42(12):757-761

D Oana Badarau conceptualized this manuscript with Eva De Clercq. Data for this paper was coded by the first author and Tenzin Wangmo, and checked by Eva De Clercq. Ms. Badarau wrote the article and all authors reviewed, commented on her work and approved the final content for publication.

(3) Katharina M Ruhe, Tenzin Wangmo, Eva De Clercq, **Domnita O Badarau**, Marc Ansari, Thomas Kühne, Felix Niggli, Bernice S Elger; Swiss Pediatric Oncology Group (2016). Putting patient participation into practice in pediatrics – Results from a qualitative study in pediatric oncology. European Journal of Pediatrics, 175(9):1147-1155

The article's topic was developed by the SNF research team composed of Tenzin Wangmo, Katharina Ruhe, D Oana Badarau and Bernice Elger. Katharina Ruhe and Tenzin Wangmo took the lead and wrote the manuscript. D Oana Badarau supported enrollment and data collection for the study, co-developed a coding map for the initial analysis of data and read, commented, reviewed and approved the final manuscript. Bernice Elger, Thomas Kühne, Felix Niggli, and Tenzin Wangmo designed the SNF study from which the paper was developed. All co-authors provided input during the writing process, reviewed and approved the final manuscript.

(4) Katharina M Ruhe, Tenzin Wangmo, **Domnita O Badarau**, Bernice S Elger, Felix Niggli (2015). Decision-making capacity of children and adolescents - suggestions for advancing the concept's implementation in pediatric healthcare. European Journal of Pediatrics, 174(6):775-782

Katharina Ruhe, Tenzin Wangmo and D Oana Badarau developed the idea of the manuscript. Ms Badarau identified key issues in defining capacity and competency from a legal and

ethical perspective. Katharina Ruhe drafted the article. D O Badarau supported the development of the legal arguments and critique. Together with all other authors, D O Badarau reviewed several drafts and approved the final article.

(5) **Domnita O Badarau**, Katharina M Ruhe, Thomas Kühne, Eva De Clercq, Anca Colita, Bernice S Elger, Tenzin Wangmo (2017). Decision-making in pediatric oncology: Views of parents and physicians in two European countries. AJOB Empirical Bioethics, 8(1):21-31

The idea for this publication was developed within the goals of the SNF study as a joint work between D O Badarau, Katharina Ruhe, Tenzin Wangmo, Eva de Clercq and Bernice Elger. D O Badarau worked on developing the analysis and themes for the paper in collaboration with Tenzin Wangmo and with the support of Eva De Clercq and Katharina Ruhe for the Swiss data. Ms. Badarau wrote the manuscript which was enriched by authors' input and comments. The final product of this work was approved by all authors.

(6) Tenzin Wangmo, Katharina M Ruhe, **Domnita O Badarau**, Thomas Kühne, Felix Niggli, Bernice S Elger; Swiss Pediatric Oncology Group (2016). Parents' and patients' experiences with pediatric oncology care in Switzerland – Satisfaction and some hurdles. Swiss Medical Weekly, 146:w14309

The idea for this article was developed by K Ruhe and T Wangmo who worked on the draft. D O Badarau contributed to the discussion section, read and provided critical input for the whole manuscript, as well as reviewed and approved the final version for submission.

(7) Katharina M Ruhe, **Domnita O Badarau**, Pierluigi Brazzola, Heinz Hengartner, Bernice S Elger, Tenzin Wangmo; Swiss Pediatric Oncology Group (2016). Participation in pediatric oncology: Views of child and adolescent patients. Psychooncology, 25(9):1036-1042

For this publication the idea was developed by the joint effort of Katharina Ruhe, Tenzin Wangmo and D Oana Badarau. Ms. Ruhe took the lead in writing the article and D O Badarau read drafts of the manuscript and provided constructive comments for the analysis, results and discussion. The final work was reviewed and approved by all collaborators.

(8) **Domnita O Badarau**, Rebecca L Nast and David M Shaw (2014): The Vulnerability of the Individual Benefit Argument. American Journal of Bioethics, 14(12):17-18

For this article D O Badarau and Rebecca Nast worked together on conceptualizing a critical analysis in reply to an article on research ethics with vulnerable populations. Ms. Badarau drafted the paper and worked closely with Ms. Nast on improving its content. Ms. Nast reviewed several versions of the manuscript and produced the final article in collaboration with D O Badarau. The manuscript was reviewed by David Shaw who commented on it and also gave permission for submission of the final article.

(9) Eva De Clercq, **Domnita O Badarau**, Katharina M Ruhe and Tenzin Wangmo (2015). Body matters: Rethinking the ethical acceptability of non-beneficial clinical research with children. Medicine, Health Care and Philosophy, 18(3):421-431

The idea for this article was conceptualized by Eva De Clercq who developed it further with the help of all authors. Eva De Clercq took the lead in writing the manuscript and coordinating the review process of all authors. D O Badarau and other co-authors commented and critically reviewed the paper and approved the final article.

(10) Katharina M Ruhe, **Domnita O Badarau**, Bernice S Elger and Tenzin Wangmo (2014): End-of-Life Decision Making in Pediatrics: Literature Review on Children's and Adolescents' Participation. AJOB Empirical Bioethics, 5(2):44-54

The literature review resulted from the four authors' collaboration on the idea for the article and design of the search strategy. K Ruhe carried out the literature search. D O Badarau and Tenzin Wangmo supported in the appraisal of resulting publications and deciding on their

exclusion or inclusion. D O Badarau was also involved in the synthesis of the final results together with the first and last authors. The final manuscript was edited and reviewed by all authors who approved the final article.

(11) **Domnita O Badarau**, Eva De Clercq, Bernice S Elger (2019). Continuous Deep Sedation and Euthanasia in Pediatrics. Does One Really Exclude the Other for Terminally Ill Patients? The Journal of Medicine and Philosophy, 44(1)

The article was conceptualized by D O Badarau in collaboration with Eva De Clercq. D O Badarau wrote the article, which Eva De Clercq reviewed and revised. All co-authors commented on the final version of the article and approved it for publication.

Study methodology

The doctoral work is part of a larger project on decision-making, competency and autonomy of children in pediatric oncology care conducted in two European countries: Switzerland and Romania. The project is a multi-center research that employed both quantitative and qualitative methods for data collection in order to achieve a comprehensive image of decision-making processes for children and adolescents suffering from cancer. Participants included in the doctoral study were enrolled in the SNSF study. Primary investigators for this project are Prof. Bernice Elger, Dr. Thomas Kühne and Dr. Felix Niggli, who also designed the study together with Dr. Tenzin Wangmo. In Romania, participants were recruited as part of the doctoral study funded by the Botnar Stiftung and the Hemmi Stiftung, Basel.

Two countries with different normative systems in relation to children's recognized right to participate in or be in charge of healthcare decision-making were selected for the purposes of the doctoral study. Swiss civil law does not make a distinction between patients' right to consent to treatment based on age, but on mental capacities. As such, minor patients who can

be proven competent should be awarded the same authority to consent to treatment as competent adult patients[1]. In Romania, parents generally have the right to make decisions for their children, as long as they are not emancipated. Parental permission and for adolescent patient's assent are customary. However, a refusal can be overruled if treatment is considered to be in the child's best interest[2-4].

Data from proxies and physicians caring for minor patients diagnosed with cancer was collected from eight of the nine independent units of the Swiss Pediatric Oncology Group and from three university-affiliated pediatric oncology centers in Romania. Among the participating centers in Switzerland, four are at university pediatric clinics and four are units within the cantonal pediatric hospitals. Participants were recruited from all Swiss regions, French, German and Italian speaking. Proxies and physicians in Romania were recruited from two distinct country regions where children from all over the country are treated for either solid tumors or oncological/hematological illnesses. For the purposes of the doctoral thesis only data obtained from qualitative interviews with proxies and pediatric oncologists in Switzerland and Romania will be presented and serve as the basis for the concluding discussion.

Data Collection

In Switzerland data collection started in August 2012 and concluded in April 2015, while in Romania it began in October 2012 and ended in August 2014. During these periods interviews and surveys were administered to physicians and proxies in Romania and Switzerland. Interviews with children were carried only in Switzerland as part of the SNSF study.

Qualitative methods

Qualitative one-on-one, in-depth interviews were used to explore the experiences of parents and oncologists when caring and making decisions related to the care of children suffering from cancer. It was also investigated if and how children (8 to 17 years old) can be included in decisions related to their care. A qualitative study design is most suited for exploring sensitive topics and seizing genuine participant experiences[5,6]. A total of 37 patient cases, 18 in Romania and 19 in Switzerland were discussed with proxies and physicians. The 63 interviews were conducted with the patients' proxies (n=37) and their treating physicians (n=26), 10 in Romania and 16 in Switzerland. Five physicians in each country discussed more than 1 patient case.

In Switzerland, interviews were also conducted with 17 minor patients as part of the SNSF study's aims and methodology. These interviews will be used for the purposes of the doctoral thesis as part of a theoretical exploration of children's involvement in research.

Quantitative methods

Interview participants were also asked to fill in one cross-sectional survey to collect demographic information about parents, physicians and patient medical data about diagnosis, prognosis, treatment. This data was used for the purposes of describing the characteristics of the participant sample in study publications.

Aditionally, as part of the SNSF project, cross-sectional surveys (n=229) gathering demographic data, diagnosis and treatment information, as well as attitudes and preferences for decision-making were completed by physicians and parents in Switzerland. In Romania, 63 surveys were completed as part of a pilot project on decision-making.

Research ethics approvals

Ethics approval was obtained from the responsible Ethics Committees (EKs) in each of the eight participating states (cantons) in Switzerland and from each Research Ethics Committee (REC) in place at the 3 collaborating pediatric centers in Romania.

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'Les grandes personnes ne comprennent jamais rien toutes seules, et c'est fatigant, pour les enfants, de toujours et toujours leur donner des explications….'

(Antoine de Saint-Exupéry**,** Le petit prince)[1]

Chapter I

Perspectives on communication challenges in pediatric cancer care

[1]Antoine de Saint-Exupéry. Le petit prince. Évreux (Eure), France; Folio, Kapp Graphic; 2015, p. 14.

Parents' challenges and physicians' tasks in disclosing cancer to children. A qualitative interview study and reflections on professional duties in pediatric oncology

Domnita Oana Badarau, Tenzin Wangmo, Katharina M Ruhe, Ingrid Miron, Anca Colita, Monica Dragomir, Jan Schildmann, Bernice S Elger.

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Abstract

Background

Professional guidelines encourage physicians to provide children with as much information regarding their health as deemed developmentally and emotionally appropriate. However, empirical research indicates that in clinical practice, an open discussion with children about cancer is often lacking. This study explores impeding factors to and possible strategies for open communication of cancer diagnosis to children from the perspectives of parents and physicians.

Procedure

Semi-structured interviews were conducted with 18 parents of children with cancer and 10 treating oncologists. The patient sample was obtained from three pediatric units in Romania. Interviews were transcribed verbatim and interpreted using thematic analysis. Inductive open-coding procedures identified participants' accounts regarding their experiences with cancer diagnosis and treatment. Final themes were selected by grouping codes that formed a pattern in the data.

Results

An interplay of mainly three different factors – information overload and emotional turmoil, lack of knowledge and skills for disclosing the diagnosis, and assumptions about burdening the child when discussing cancer – restricted parent-patient communication and subsequently affected physician-patient exchanges. Oncologists recommended open communication at diagnosis, but left the final decision to the parents. They adapted their communication style with patients to parents' preference.

Conclusions

Although physicians need to respect the wishes of children's legal representatives, they also have a duty to promote patients' best interests. We recommend that physicians employ a proactive stance in ensuring that children with cancer are appropriately informed about their diagnosis. In case of parents' arduous objections to full disclosure, an ethical consultation should be considered.

Introduction

Professional guidelines recommend that communication of diagnosis for children with cancer should be conducted in a planned manner, and include both the patient and the parents. Physicians are encouraged to provide children with as much information as deemed developmentally and emotionally appropriate. At the same time, communication should take into account parental wishes[1-5]. The recommendations on communication of diagnosis to minor patients are supported by a growing research body evidencing the positive outcomes of communication with children and adolescents. Quantitative and qualitative studies show that children who are informed gain a sense of control that not only reduces anxiety and facilitates treatment, but also helps them understand their illness[6-8]. However, these studies were conducted in different settings, such as emergency department, surgery, infectious diseases, and oncology, and not all included children with life-threatening illnesses. There is a lack of studies capturing the benefits of open communication for children with cancer. One study found that children wished to be informed to different degrees, with some wanting to know the bare minimum[6]. In another study, although parents recognized the advantages of open communication, they expressed difficulties with child's presence particularly when sensitive information, such as prognosis, was discussed[9].

Empirical studies on the practice of communicating diagnosis of cancer with children indicate that there often is a considerable gap between recommended and actual practice. Parental control on both the content and completeness of information provided to children and the desire to protect them from distressing news have been identified as contributing factors to this gap[8,10-15]. However, there is scarcity of research on how to deal with situations in which physicians' open approach to diagnosis communication conflicts with parental behavior aimed at avoiding direct communication.

This study provides a detailed reconstruction of the communication of cancer diagnosis to children in Romania by means of semi-structured interviews with their proxies and treating oncologists. The focus of the analysis is on the identification of factors that contribute to restricted provision of information about diagnosis to children. Based on the empirical findings, we discuss potential strategies and professional duties to reach an open communication with children with cancer in cases when parents oppose disclosure.

Methods

Conducted in Romania, the study is part of a larger project on children's participation and decision-making in pediatric oncology. It is based on semi-structured interviews carried out with physicians and parents of minor patients diagnosed with cancer and undergoing treatment in three university-affiliated pediatric centers. We used purposive strategy to sample participants[16] based on the following criteria: they are the parents or treating physician of a minor patient aged 8-18 years receiving cancer treatment at one of the study centers. When parents were not the main caregivers, the proxies accompanying patients were approached. The treating physicians selected patient cases fulfilling the criteria. Proxies judged by physicians unsuitable for enrollment due to child's delicate situation or burdening psychosocial difficulties were excluded. In total, 21 proxies were introduced to the researcher for a detailed discussion about the study's goals and procedures and from these, 18 participated in the study. Researchers also invited the treating physician for each corresponding patient case to be interviewed. Proxies' and physicians' characteristics are described in detail in Table I. Participant recruitment was carried out between May and October 2013, and all interviews were conducted in person at a time deemed convenient by each participant. Informed consent was obtained from all participants. The study received

approval from the Ethics Committee at the researchers' institution in Switzerland and the institutional Ethics Committees at every study center.

Table I. Characteristics of proxies and physicians

Proxies	**(n=18)**
Male	1
Female	17
Age[a]	
34-39	7
40-45	4
> 46	5
NA	2
Months since child's diagnosis	
0-6	8
6-12	8
> 12	2
Physicians	**(n=10)**
Age	
30-39	3
40-45	3
≥46	4
Experience (years)	
0-4	1
5-12	5
>12	4

[a] Data missing for two proxies

Interview questions were semi-structured according to protocol in three categories (Fig. 1) and allowed participants to freely describe their experiences. Whenever necessary, the interviewer asked prompting questions to explore topics generated by participants[17,18].

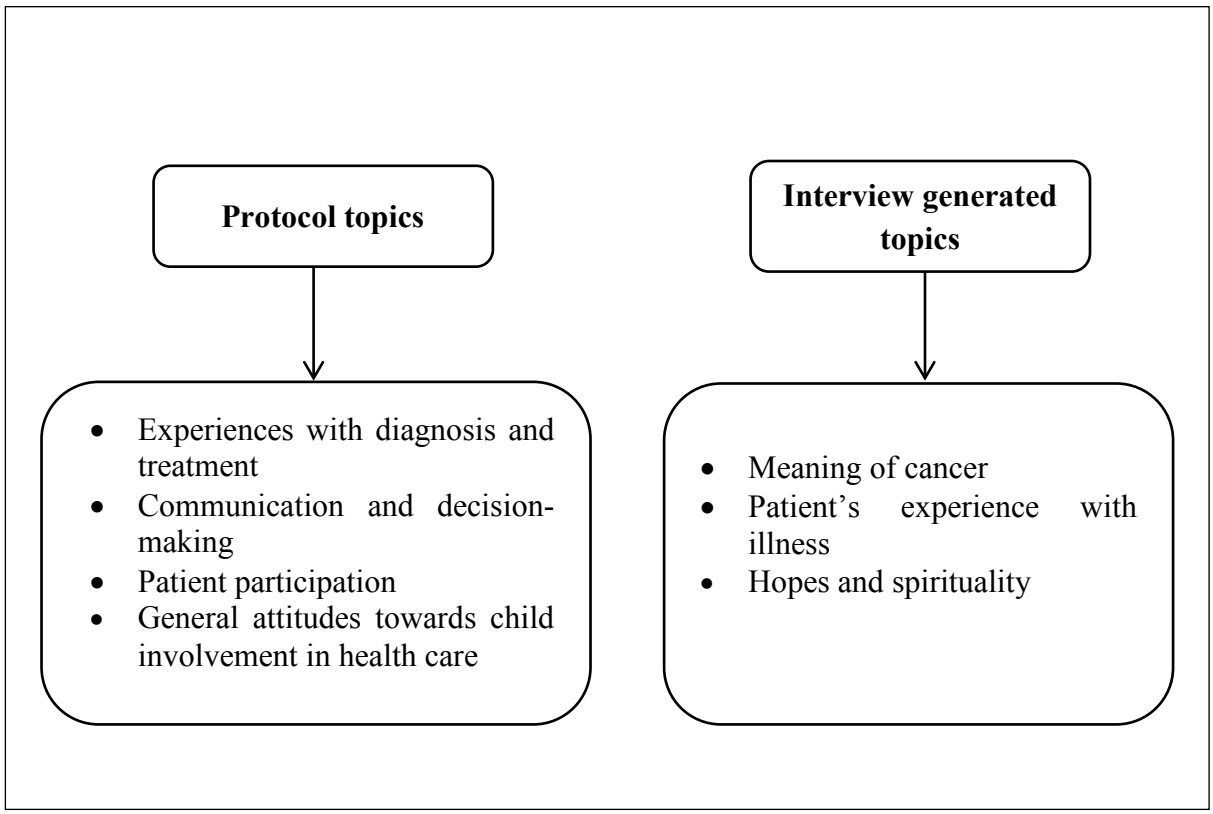

Fig. 1. Interview topics

All interviews were analyzed using thematic analysis[19] to identify and select accounts reported by participants when talking about their experiences of caring for a child with cancer. In a preparatory phase, three researchers (D.O.B., T.W., and K.R.) forming the core research team and trained in qualitative research methods used inductive open coding for a sample of three interviews. This initial coding was done independently and codes were compared and discussed among the team until consensus on a systematic coding scheme was reached[19]. All interviews were then analyzed inductively following this coding scheme. For any emerging issues described by participants, new codes were added. Thereafter, codes identified to form a pattern were grouped under specific themes that were discussed within the research group. Finally, we selected specific themes relating to the communication of diagnosis topic. The selected themes and coded extracts of data pertaining to these were reviewed together with an independent researcher familiarized with the interview data (J.S.)

and further refined. We discuss the product of this final analysis in the Results section and provide typical quotes from the original interviews as illustration.

Table II. Patient information

ICCC-3 main classification diagnosis[a]		Patient[b]	Age group[c]	Sex
I	Leukemias	Chris	I	M
		Cody Ray	I	M
		Aaron	II	M
		Bruce	III	M
		Melissa	I	F
II	Lymphomas	Anthony	III	M
		John	III	M
III	CNS and miscellaneous intracranial and intraspinal neoplasms	Tracy	I	F
		Celine	II	F
		Carrie	II	F
		Harry	I	M
		Rick	II	M
		Sawyer	II	M
IX	Soft tissue and other extraosseous sarcomas	James	I	M
		Tyler	III	M
X	Germ cell tumors	Desiree	I	F
XI	Other malignant epithelial neoplasms and malignant melanomas	Steven	II	M
		Lisa	III	F

M, male; F, female;

[a]International Classification of Childhood Cancer, Third Edition (ICCC-3). http://seer.cancer.gov/iccc/iccc3.html;

[b]All patients have been assigned a false name following the data anonymization process;

[c]Age group I (8-12 years), age group II (13-15 years), age group III (>16 years).

Results

In total, 28 interviews were conducted with proxies and physicians of 18 patients aged between 8 and 18 years. Patient characteristics are described in detail in Table II. From the 21 proxies approached, one mother declined participation, two others were not available for interviewing due to patient's condition, and 18 consented to participation. The participants in the proxies group included 14 mothers, one father, and three grandmothers. All 10 physicians approached and caring for those 18 patients participated, with five physicians being interviewed for more than one patient. Twenty-four interviews were recorded and for three, the interviewees (all proxies) declined the use of audio devices. One more interview was partially recorded as the participant was overwhelmed by emotions. To allow the participant to recompose, the researcher stopped the device and out of sensitivity, when the participant continued to tell about her experience, did not restart it. Recorded interviews were translated and transcribed in English. Detailed notes were taken for the interviews that had not been (completely) recorded. On average, interviews with parents were 53 min long (range: 25 min- 1 hr and 20 min), whereas physician interviews lasted around 50 min (range: 26 min- 2 hr and 3 min).

Based on our analysis, the common model of initial communication of diagnosis was a discussion between parents and physician only. According to physicians, this discussion was usually well planned with the aim to explain the diagnosis and following procedures. Physicians perceived this discussion as a starting point for a process of communication about disease, prognosis, and treatment options and also as a means to get to know the family.

> Desiree's physician: '[…] First we talk with the parents, with the surrogates, who are accompanying them. […] Afterwards, with time...ahhh...you cannot know, neither the surrogates, nor the child from just one talk. Usually you

know them along the way. You know them, their character, temper, this psychological side.' (Patient age 11)

Factors limiting communication about diagnosis between parents and children

The narratives of parents about the time subsequent to the first discussion with the physician indicate that some parents took a proactive role in informing their children about their disease. However, several parents reported that they did not or only partially inform their children about the disease. One factor reported by parents as contributing to a restricted or lack of communication with their child was information overload and emotional turmoil. Communicating the diagnosis to their children was broken down to several continuous, unstructured stages. Parents reported explaining the illness at first and gradually starting to provide more specific information, such as naming it and filling in the blanks for their children (Quotes 1 and 2 in Table III).

Parents identified lack of knowledge or inexperience with the medical setting to be another inhibitor to open discussions with children. These parents described being unsure about how to have such a first talk with their child and feeling unprepared for discussing the cancer diagnosis (Quote 3 in Table III).

Table III. Quotes from interviews with proxies

Number		Quote
1	Desiree's mother (Patient age 11)	'We didn't even have time to process [the diagnosis information] very well...what it is happening. […] she didn't know the "cancer" term. She knew that she had a booboo. For a while we said cyst... I couldn't pronounce the word tumor. I wasn't ready….'
2	Tyler's mother (Patient age 17)	'They did the biopsy. Bad news! It is a tumor (*speaking slowly*). […] And so the entire craziness started, the treatments. […] So it was horrible. […] Ah, what I told him .. Well, I could not tell him so directly (*talking very slowly*). I only told him that he has a problem there and that he will be OK, that he will get well with the treatment.'
3	Aaron's mother (Patient age 14)	'Afterwards... I did not know how to tell him (Aaron)…how to inform him and to … [tell him] this ... that he has leukemia. He goes to the play room, upstairs, they gave him those ... magazines with "fight cancer" this and that and he ... nothing ... would throw them away ... the papers, the magazines ...'
4	Cody Ray's mother (Patient age 10)	'I avoided the topic, you know? But I realized that if he had heard about this diagnosis, he would have been very traumatized and scared and even ... Well, you do not know how he would react.'
5	Carrie's grandmother (Patient age 14)	'It is hard for me. I cannot, I cannot tell her because she knows how dangerous this illness is. She knows it from her mother [who died of a brain tumor]. And I cannot make her worry and make her say: "god, I also have the same thing. And maybe tomorrow, the day after tomorrow..." No, I cannot.'

A third factor identified as a barrier toward discussing the diagnosis was the assumptions that communication about the disease would be a burden to the child. Some parents expressed incapacity to fully reveal the diagnosis due to uncertainty regarding how children's attitudes toward the illness might change. Others anticipated an emotional harm whose impact they found difficult to assess or control. In part, these expectations were influenced by the fact that parents (and patients) had been in close contact with (other) family members suffering from cancer. Particularly, parents feared addressing the life-threatening nature of the illness. They did not want children to make a connection between their diagnosis and the possibility of dying from it (Quotes 4 and 5 in Table III).

Physicians' perceptions and strategies in cases of lack of open communication

Interviews with treating physicians revealed that they were often aware of the state of communication between parents and children and respective deficits. In such cases, some physicians reported that they had recommended an open communication. However, physicians mentioned that they did not directly interfere in the communication within the family and left to the parents the control over whether they disclosed the diagnosis to patients (Quotes 6 and 7 in Table IV).

However, in cases where parents did not openly communicate the diagnosis to their children physicians said they talked to the patients themselves. They described several communication strategies when discussing directly with the child. One of these strategies was to stay focused on factual medical information in talking to the patient, while maintaining an "air of optimism" (Quote 8 in Table IV).

Table IV. Quotes from interviews with oncologists

Number		Quote
6	Harry's, Sawyer's and Celine's physician (Patients ages 11, 15 and 13)	'I also ask the parents. Many times the parents tell us to not tell them [children] because he [patient] does not know what he has. And then I am the one who insists: "But what does he know that he has? He must know something. He is coming from the physician, he had surgery"'.
7	Aaron's physician (Patient age 14)	'So, we wish to tell them [children], but if the parents tell us that under no circumstances they cannot know, we respect this wish.'
8	Harry's, Sawyer's and Celine's physician (Patients ages 11, 15 and 13)	**'**He [Sawyer] knew that he had a tumor there, that he had surgery, that there is still some residual tumor and that we have to treat the rest. We did not go into details and it did not make sense to go into details like: malignant tumor, or other tumor... Plus, pilocytic astrocytoma sounds so good. On the other hand, we do not lie too much about it because fortunately pilocytic astrocytoma is considered benign, but unfortunately it can have a poor evolution and can behave like a malignant tumor.'
9	Cody Ray's physician (Patient age 10)	'If the patient [...] comes to me and asks me specific questions, I would say the truth. But I let them [patients] make the first step. I do not want to tell them myself directly because many times this (the whole experience) does not end up well.'

Physicians perceived parental approach to limit communication as problematic. As legal requirements in Romania give parents authority over healthcare decisions for their minor children, physicians perceived a conflict between the responsibility to inform patients truthfully and the desire of parents to withhold information. As a consequence, most physicians reported to adopt a non-direct and passive disclosure strategy. Several physicians said they preferred to only react to patient inquiries and explicit concerns. Their descriptions of how they would react if directly confronted by patients to answer the diagnosis question showed clearly that they were unwilling to deceive (Quote 9 in Table IV).

Discussion

Communication of diagnosis in pediatric oncology is a complex process determined by triadic influences on the direction and depth of communication[14,20]. Our study has the advantage of confronting both parents' and physicians' views on disclosing the diagnosis of cancer to children. The qualitative design of the study facilitated obtaining in depth information on factors hindering parents from open communication with their child and physicians' reluctance to intervene in such situations. This provides new insights into diagnosis disclosure practices and points to strategies for facilitating an open communication with patients. In the discussion, we use these empirical findings to explore feasible procedures to improve communication of cancer diagnosis and professional duties in cases of lack of open discussions between parents and children.

Our research indicates that an interplay of mainly three different factors – (1) information overload and emotional turmoil, (2) lack of knowledge and skills with disclosing the diagnosis, and (3) assumptions about burdening the child when discussing cancer - contributes to restricted communication between parents and children. Parents of children diagnosed with cancer consider the presence of minors in discussions of diagnosis

problematic[9] and sometimes resist disclosing the illness to their children[8,20,21]. Our study shows that, in spite of cultural differences, these attitudes are more likely to be affect-driven (1). It may come as a natural parental response to a life-threatening situation that physicians should be aware of. Participants in both groups acknowledged the great impact emotions had on communication of diagnosis, as well as on the content of information provided to children. The more troubled the parents were, the more difficult it was for them and physicians to give the patient complete diagnosis related explanations. An inclination toward limited sharing of information may be stronger at the time of diagnosis and shortly afterwards. Similar to a study conducted only with parents[9], participants reported the struggles proxies faced and their avoidance of discussing the life-threatening aspect of the illness. However, in our study, informing patients about side-effects was not as problematic, as physicians would take the lead and support proxies in explaining and re-assuring patients.

At the same time, this study suggests that physicians need to be trained on how to explore different reasons that prevent parents from communicating openly and to select appropriate strategies. In this respect, physicians need to know for example how to deal with parents' information overload and emotional turmoil. Proxies in our study reported an acute need to be listened to and to have several detailed illness related discussions with physicians at the time of diagnosis. Although this need was identified by oncologists in most cases, they felt pressured by lack of time. They considered a solution the advice they provided to parents to be open with children about the illness.

Other studies[10,22,23] have shown that in some cases, it may be sufficient to let time pass to allow parents to cope with the cancer diagnosis and enable them subsequently to speak openly to their children. In other cases, additional information for parents on the adverse outcomes of limiting information or consultation with a member of the psychosocial team may be required. Furthermore, physicians have to understand that it is normal that parents

lack skills to communicate a life-threatening diagnosis (2) and they routinely need help. Although in some countries, large medical centers have a communication of diagnosis protocol requiring multidisciplinary support (usually a social worker, nurse, psychologist) for parents and children for the initial discussion sessions[24-26], this is not the standard in many countries and even not for all institutions in countries with well-developed healthcare services. Communication of diagnosis is not always conducted by a multidisciplinary team[10,22,27] and it may happen during routine check-ups, in the emergency, or surgery department[10]. This was the case for the oncology units where our study was carried. Child psychologists to whom patients can be referred were present, but a coordinated care system lacked. A multi-professional team with specialist nurses and psychosocial trained members to support parents were not in place in these institutions. However, volunteering social workers were present at all participating sites and psychosocial care offered by social workers was available on specific week days. These services were offered through a large national privately funded charity (P.A.V.E.L. Association)[28]. At the same time, it should be acknowledged that parents vary in their preferences regarding who should be present for the communication of diagnosis[25].

Physicians, nurses, or even other parents with similar experiences can provide support by suggesting "how" to discuss bad news with a child, particularly when there is no multidisciplinary team approach. After all, this is a difficult task also for professionals[29-31] and it seems likely that elements of open communication can be learnt more efficiently by parents with healthcare professionals' aid[27,32]. Physicians also need to be aware that reported anticipation of possible harm to the child (3) requires them to further explore parents' reasons. Physicians should routinely discuss with parents the benefits and harms of an open versus restricted parent-child or physician-patient communication. During the communication

of diagnosis discussion with parents and subsequent encounters, physicians could offer to disclose information to children together with parents.

Our study also clearly points out a need for further training of physicians with regard to ethical and legal issues involved in (non-)disclosure of cancer diagnosis to children. One such tool for training is the Oncotalk[33], a teaching module to improve communication skills. This is freely available online (http://depts.washington.edu/oncotalk/) and can be a great source of developing skills for breaking bad news. It is available only in English, which may sometimes be a barrier for some healthcare professionals in different countries. In our study, data indicate that physicians do not actively engage with or sufficiently support parents who repeatedly object direct communication to carry out more open discussions with their child. Using the Oncotalk tool, which includes a module on teaching family conferences, may help overcome this. Moreover, physicians in our sample seem to be wary of what is the most appropriate way to deal with legal provisions that entitle the child's proxy to set the tone for physician-patient discussions. Balancing this requirement with patient's best interest when it may go against parents' wishes is a delicate issue. Physicians' reluctance to challenge parents may be tied to considerations for the deep bond parent and child share, but also to apprehension of litigation[34]. Accordingly, the question from a professional and legal point of view is what should physicians be expected to do in situations when there is no open discussion with the child about cancer? Although legal aspects with regard to parental rights must be considered[2,21,35,36], we argue that physicians underplay their duties to act in the best interest of children, as studies show a beneficial effect of information on some children and negative effects for lack thereof in others[6,7]. From an ethical perspective, such circumstances demand a proactive stance on the physicians' side to elicit patients' own preferences regarding information provision, which can be defensible even from a legal point of view[35]. Although it is beyond this article to provide a concrete operating procedure, we suggest that a

first step is to schedule regular follow-up meetings with parents to discuss the state of information between parents and children. Furthermore, these discussions between physicians and parents should include an exploration of possible factors contributing to parents filtering information or employing avoidance techniques when discussing disease and treatment with their children. Identifying hindering factors leads to a better understanding of parents' attitudes. At the same time, it provides physicians with an opportunity to address them and to point out the potential harms, such as isolation and anxiety, that children face due to lack of clear communication[6,7]. It is important that physicians remain understanding toward parental attitudes and avoid coercion. Instead, they should employ techniques for encouraging and supporting parents in open communication to children[21,35,37]. In cases when even a repeated proactive approach by physicians and the intervention of psychological support fails to produce an open communication with the child patient, we suggest to use ethical case consultations or comparable interventions. This intervention should be sought primarily when resources within the team are not sufficient to deal with the situation at stake. As such, it is rather complementary and its focus should include context sensitive analysis of all clinical, personal and ethical issues in question. Additionally, it should involve and engage with all members of the medical team and family.

Limitations

There are a few limitations with regard to this research. First, this study was conducted in three Romanian centers, and therefore findings should be interpreted against the background of cultural and center-specific organizational factors. However, there is no reason to believe that the included centers have unique characteristics or adopt special attitudes as compared to other centers in Romania. Although our data were collected in one country, as pointed out in the discussion, we identified a number of characteristics of communication between parents,

children with cancer, and physicians that seem rather typical for the clinical pediatric oncology setting and go beyond the selected country and cancer centers. A second limitation is that we recorded participants' experiences within different lengths of time from diagnosis. Re-call bias may have influenced participants' accounts. Interviewees' answers may have also been biased by social desirability. However, in light of the honest accounts of partial disclosures of diagnosis information to patients as reported by both participant groups, this bias seems to be modest. A bias may be also caused by the selection process as we relied on collaborating physicians' interest in the study's topic and their drafting of patient cases. Finally, although the design of our study allowed matching the narratives of parents and physicians, we did not elicit the perceptions and views of the children themselves. Therefore, research which includes patients' perspectives may yield further information that is relevant to guide parents and clinicians with regard to communication with minor patients about cancer.

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Cancer care in Romania: challenges and pitfalls of children's and adolescents' multifaceted involvement

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Abstract

Communication about diagnosis and medical treatment for children suffering from life-threatening illnesses is complex. It is a primary step in involving underage patients and families in care and lays the foundation for obtaining parental permission and patient assent for treatment. In practice child participation in care is often difficult to obtain due to patients' different and sometimes fluctuating preferences, but also parents' protective strategies. Physicians may be susceptible to parental wishes to limit information and feel uncomfortable discussing issues related to uncertainty of cure with patients. A qualitative study in Romanian pediatric oncology units was conducted to explore children's involvement from the perspectives of parents and oncologists. Interviews with participants discussed 18 patient cases. Data were transcribed and thematic analysis was used to interpret and mine patients' involvement during treatment. Different facets of patient participation were identified: restricting, widening and enhancing involvement. A fourth category, unintentional involvement, occurred for all patients due to children's observations during long-term hospitalizations and access to Internet. Uncertainty overarched parental attitudes regarding the extent to which children should be included. Physicians usually complied with parental wishes to limit involvement, but together with parents involved patients at least in a practical way. Adults' protective attitude may backfire, as adolescents' online searches often expose patients to worse-case scenarios. Further research should acknowledge the hazards of restricted diagnosis disclosure and develop clinician tools to support families in communicating with patients. This should be paralleled by physician efforts to elicit patients' needs regarding participation.

Introduction

Provision of information is a fundamental step towards shared decision making. It enables minor patients and families to frame personal values when making decisions[1]. Communication is a complex process and studies report that children wish to know what is happening to them and for clinicians to listen to their preferences[2-4]. Some patients prefer to know only certain aspects, to receive information from their parents or to leave decisions up to adults[3]. The process is further complicated by parents' reaction to diagnosis and inclination to protect children from distressing news[5,6]. Most physicians consider it disrespectful to withhold information, but may comply with parental restrictions[7].

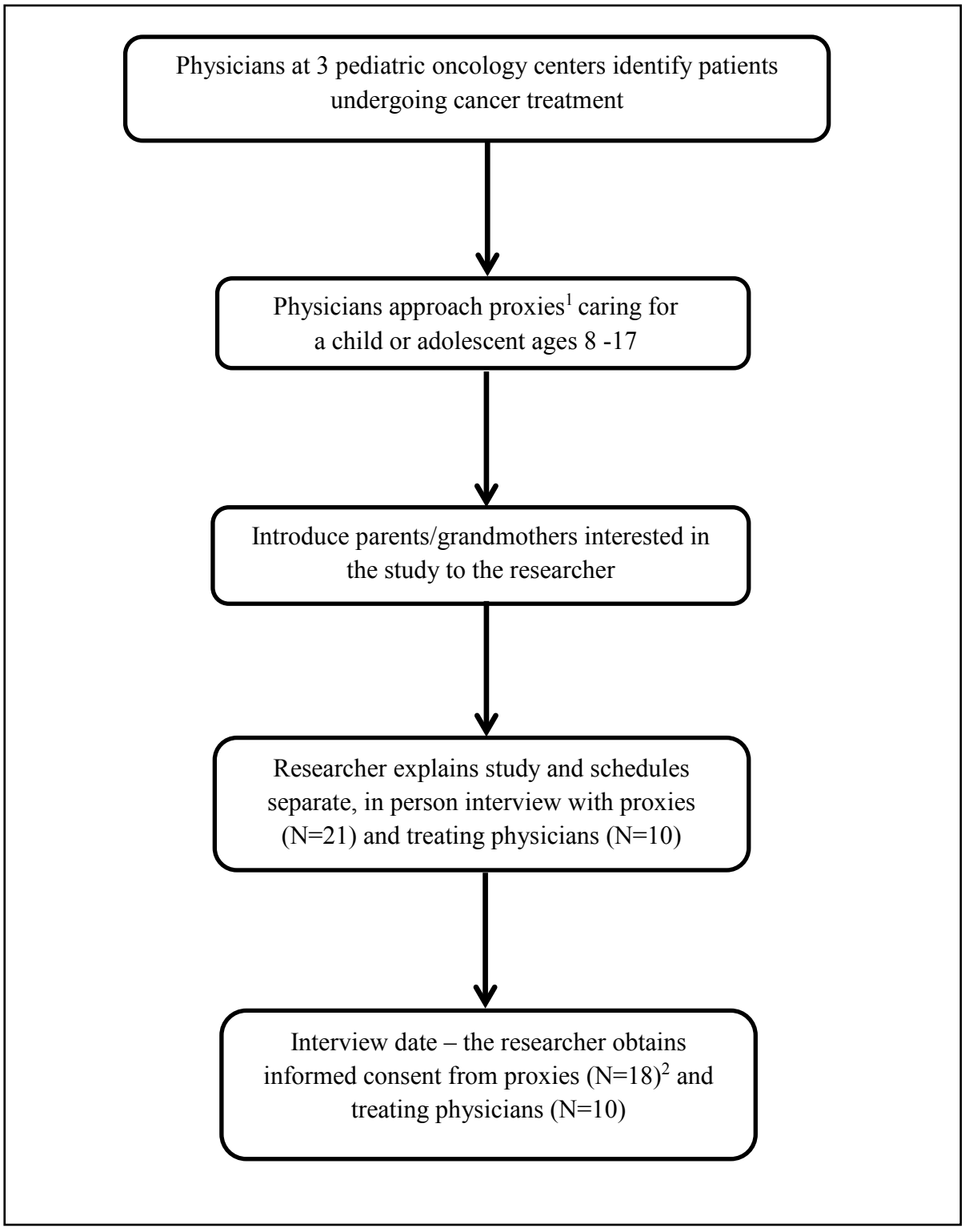
This study is part of a project conducted in Swiss and Romanian pediatric departments. It explores the complex process of involving patients with cancer in their care from the perspectives of oncologists and parents in Romania. It analyzes how adults act either to limit or support patient participation by evaluating the impact of these actions on children[i].

Methods

Semi-structured interviews were conducted with proxies and treating physicians of patients aged 8-17 receiving treatment at collaborating centers in Romania. Interviews with children were conducted only in Switzerland and are reported elsewhere[8]. We employed purposive sampling[9] to enroll 28 non-representative participants between May and October 2013 (Figure 1). The study was approved by the ethics committee at the researchers' home institution in Switzerland and institutional ethics committees at Romanian participating centers. Informed consent was obtained from all participants.

[i] When used in text, the word child is meant to represent all minor patients, including adolescents, unless otherwise specified.

Figure 1 Participant recruitment procedure



[1] Proxies - parents or caregivers with legal power. Three patients were accompanied by grandmothers acting as proxies and in charge of care; all other 15 interviews were with parents.

[2] 3 proxies declined participation due to child's advanced illness or disinterest in the study's topic.

Interviews were recorded, transcribed and thematic analysis was applied for all datasets[8] by identifying codes that formed a pattern and organizing them in major themes synthesizing participants' experiences[10].

Results

Eighteen proxies and 10 physicians of 18 patients participated in the study[8]. Parents' and physicians' attitudes contributed to the pace of involvement which can be construed by four themes: restricting, unintentional, widening and enhancing involvement. The unintentional category applied to all patients. Children either passed through all involvement stages, from restricting to enhancing, remained in the restricted and unintentional categories or started from limited involvement at diagnosis and leapt into the widening or enhancing phase.

Restricting involvement

Many physicians reported that at diagnosis time they often refrain from disclosing the exact nature of the illness to patients due to uncertainty about parents' preferences on how much children should know. Clinicians found it challenging disrespecting parents' attitudes on information provision and accepted to restrict involvement at parents' request. This impacted the timing and depth of some patients' involvement and limited information provided at diagnosis and shortly afterwards. (Quotes 1 and 2 in Table 1)

Parents requested partial disclosure because of fear that the word "cancer", usually associated with dying, would have a negative impact on their child's willingness to fight. (Quote 3 in Table 1) Retrospectively, parents regretted and recognized that they cannot shield children from all distressing information and the steps they took may not entirely control what children, particularly teenagers learnt about the illness. (Quote 4 in Table 1)

Table 1. Restricting child and adolescent involvement

	Participant	Quote
1	John's physician (Patient age 16)	'[…] during the first discussion, we do not know exactly what the parents want us to do, if they want the child to find out about it [diagnosis] and how they want the child to find out. And so we respect the parents' decision. […] If the parent tells us from the beginning: "He does not know and we do not want him to know..." in that case, we also have to be careful what we talk with the parents when the child is present.'
2	James's father (Patient age 11)	'I said... that I don't want the boy to think that this is an illness (without hope) […] I said: "It is a tumor, sweetie". […] He (the physician) did not tell him about the illness, because I told him (James) that he has a tumor and it was not necessary for the physician to tell him too.'
3	Cody Ray's mother (Patient age10)	'[…] I hope that he will not make the connection (that leukemia is cancer of the blood). […] I saw that he was scared of this word "cancer". You know what I mean? That is why I am scared. I am not scared about something else. So he knows that you die from cancer.'
4	Melissa's mother (Patient age 10)	'You do not have time to think about these things before. They happen to you and you act instinctively as a result and you just go ahead. She asked me if leukemia means cancer and I told her that it does not. I lied to her about this. […]I really regret that I lied to her but to be honest, how can you tell this to your child?'

Unintentional involvement

Despite desires to control the pace of information provision, parents could not buffer the outside world or prevent children from seeking information from other sources, such as conversations on the ward and the Internet. Both parents and physicians were aware that parental boundaries on children's knowledge about their illness were fragile and could easily be transgressed. (Quotes 5-7 in Table 2)

Involvement was in many ways unavoidable in the context of hospitalization. Physicians mentioned that children were exposed to observation during medical rounds, other patients' experiences, and medical procedures. This also implied that patients were confronted with the possibility of treatment failure and of dying by witnessing other patients' evolution. (Quotes 8-10 in Table 2) Some physicians mentioned that they did not want to take the first steps towards revealing the exact diagnosis. This way those children who wanted to know may decide whether to seek more information, how much and when. (Quote 11 in Table 2)

Widening involvement

As parents and patients became more acculturated with hospitalizations and had the time to come to terms with their new situation, many parents slowly abandoned efforts to shield children. While some parents involved their children concurrently or immediately after they were told the diagnosis, for most parents the gradual awareness that involvement was somehow inevitable made them change their attitude and support widening child participation. Over time they provided more information in steps. Parental avoidance of full diagnosis disclosure persisted only for few patients after treatment started. (Quotes 12-14 in Table 3)

Table 2. Unintentional child and adolescent involvement

	Participant	Quote
5	Cody Ray's mother (Patient age 10)	'[…] he also knows he has leukemia. He found out […] From parents who have younger children and their children do not understand what it is happening and they talk about it without thinking about it.'
6	Lisa's grandmother (Patient age 17)	'But after they [physicians] gave us all (reports) from the biopsy, she then took the paper and looked it up (on the Internet). Well, she found it (medical forms). I could not hide it from her anymore. […] And so she started to look it up online. How it manifests, how it can be treated and all sort of... all these things, other cases.'
7	John's physician (Patient age 16)	'[…] in the end, they find out. It is impossible that they do not get their hands on the discharge forms, even if it does not happen after the first hospital admission. And after that they look it up online and they know.'
8	Melissa's physician (Patient age10)	'I think that nevertheless, they know....They hear about it. It is impossible for them not to hear about it. They hear what treatment they are getting and they kind of know the name of those substances.'
9	John's physician (Patient age 16)	'But in general, the children find out. And I think it is better that they find out from the healthcare workers, best suited for this is the treating physician, so better from him than from (reading it up on) the Internet or from other children.'
10	Cody Ray's physician (Patient age 10)	'[…] after two-three rounds when obviously they found out about it. They know because they come for the treatment, their hair falls out. […] [To do it like this] For them to find out on their own and afterwards for them to learn more from me. For me to see how much they want to know from me.'

For many patients widening involvement mainly consisted of information on treatment and possible side-effects. Parents and physicians considered these discussions necessary to prepare children and ascertain their collaboration for painful procedures. Physicians often reported that they could not force children to undergo treatment and some parents acknowledged child benefits from participation. Typical of this "practical" involvement is that when parents resisted open communication about the diagnosis it did not always lead to full disclosure. This resulted in physicians and parents walking a fine line between providing and withholding information. (Quotes 15-17 in Table 3)

Enhancing involvement

Physicians described children's involvement in care in relation to the information they provided and to actions taken to elicit preferences and give children a voice. (Quotes 18 and 19 in Table 4) Enhancing involvement was grounded in assessing patient perceptions and tailoring information to expressed needs[11]. According to physicians, some children limited themselves to asking more daily-life affecting questions. Other patients were curious and took on a more active role regarding the course of the illness, recommendations for treatment and dietary regimens. (Quotes 20 and 21 in Table 4)

Parents also mentioned discussing with physicians test results and treatment uncertainties in children's presence. This tactic allowed patients to participate in the sharing of information and created opportunities for them to react. Proactive parental approaches helped support and encourage some children to ask questions and engage with them regarding what patients learned from observation or other sources of information. (Quotes 22 and 23 in Table 4)

Table 3. Widening child and adolescent involvement

Participant		Quote
11	Tyler's mother (Patient age 17)	'[…]. I had a copy (of the discharge form), and he asked: "Give it to me too so I can look at it". I cannot tell him... He is older. I cannot hide it from him anymore, no matter how you try. And even if I for instance would hide it from him, he will still find a document at home, no?'
12	Harry's mother (Patient age 11)	'The physician there (told us). Yeah, it was just me and him (the son). […] My boy knew from the beginning all that he has because we did not hide it. Because we thought that if we hide from him, it might be worse for him later.'
13	Aaron's mother (Patient age 14)	'Like I said, in the beginning I would hide things, but now I do not have things to hide anymore because I would cause him more harm because he understands that...what he still has to endure, he has to go forward.'
14	Lisa's physician (Patient age17)	'I have to tell them something, no? […] you have to explain to each one of them, because there is one thing to have to stay two weeks at home and another thing is to stay just a few days, a week maximum. And of course, it is good that they are informed and that... they know about it. Of course, I explained to her what chemotherapy is, what she has to do, daily, how many days she has to stay in the hospital, around what time she can be released, what to expect after the treatment. […]'
15	Desiree's mother (Patient age 11)	'I wanted to always tell her what's next slowly by slowly. […] I always told her "look, next is...". I never took her by surprise. Next we have to...we were told "look, we start a treatment. It is called chemotherapy. We put a certain substance of anti(something)", so that she will not be sick, with nausea. […] At some point, there was nothing, nothing to be hidden from her. […] at some point the talks are somehow... in the open, like this, I don't know. No. It is better if she knows and she is also safer when she also participates there.'
16	Tyler's physician (Patient age 17)	'Even if I don't communicate the [exact] diagnosis to the child, actually you also have to get consent from the child. And I always explain to the child: "You know you have some... It hurts you... it must stop hurting you, right?" You have to explain it in a way that he also realizes that he also wants it (the treatment), that is, that he also agrees to have this treatment within his limits of understanding.'

Table 4. Enhancing child and adolescent involvement

Participant		Quote
17	Harry's physician (Patient age 11)	'I encourage them and try to, so to say "make them", I insist that they ask me absolutely everything that they want to.'
18	Lisa's physician (Patient age 17)	'Basically I talk with the patient every day. And when we talk every day, some questions that might interest them about various things, like the evolution and so forth, come up.'
19	Tyler's mother (Patient age 17)	'When we got discharged […] The physician asked us to give him an email address. And then, I sent Tyler (to talk to the physician and give the e-mail address). […] I said to him: "The physician said that you should go to him to give him your email address so that he can communicate with you directly".'
20	Desiree's mother (Patient age11)	'But she even goes and asks about the test results. She learned which are the white cells, what do they mean, the neutrophils, she gets involved and she is calm if she knows. […] She already knows...I have low neutrophiles I am not allowed outside, I have no immunity.'
21	Lisa's physician (Patient age 17)	'The girl is also present for the discussions because she is older, she is open, she is smart. She knows what she has. And she also has a lot of questions to ask. So she also wants to get explanations.'
22	Bruce's physician (Patient age 17)	'He is not a very, very curious adolescent. […] In general he wants to know how many days is the treatment session, if it is with methotrexate or without, because he knows he will get mouth sores.'

Discussion

Communication about cancer is shaped by societal views of illness and breaking bad news practices[12,13]. This study adds knowledge on the challenges and risks of involving children in care when they are not immediately or fully informed about their cancer diagnosis.

Patient participation was described as a step-by-step, intermittent process resulting in various types of involvement. Provision of information and explanations regarding care are the main tools to foster participation in pediatric oncology[14,15]. In our study, participation was dominated by parental and physician uncertainties regarding how and to what extent they can and should involve their children[3,7]. Uncertainty was linked to fears of children's emotional reaction to the cancer diagnosis. For parents themselves the diagnosis evoked a powerful threat, of dying, which accounts for the withholding techniques[16].

Physicians often complied with parents' requests to withhold the exact diagnosis in view of legal parental rights, but most saw it merely as delaying patients from finding out on their own[3]. This results in an ethical conundrum: limiting truth telling poses moral internal dilemmas for physicians, leading to conflict with some parents[7]. Despite uneasiness, physicians in this study complied with parental wishes. This does not reflect a denial of patients' right to information, as patients were gradually involved in care, even in the absence of full disclosure for some. This finding highlights the grey zone of how to professionally and empathically disclose diagnosis to families and children[17]. The degree of involvement desired by patients is difficult to assess especially when parents and oncologists take an adult-centric view and exclude child participation from what they consider difficult situations[2,18].

Participation and information sharing related to treatment occurred even as physicians accepted parental restrictions. Physicians considered discussing likely side-effects and explaining treatment procedures an imperative as these often result in pain and considerable physical changes. This communication was a way of soliciting and obtaining assent. As such,

children's involvement was seen as valuable in and of itself, rather than just a foreseeable maneuver to achieve patient compliance and collaboration. In other studies, clinicians held the same view that causing harm to children is ethically charged[7,19].

For some patients, communication became more open between parties and information flow was less and less inhibited by parental or physicians' attitudes. Oncologists allocated time for children to ask questions and reassured them with each medical visit that they could voice concerns. These actions aimed to build patient confidence to engage in care and facilitate a two-way communication. Similarly, parents supported children by including them in discussions with physicians and in some cases encourage them to have direct contact with clinicians. These attitudes may be highly supportive of adolescents' needs during cancer care[3,4]. In early diagnosis stages, children may prefer parents to act as buffers and messengers of medical communication[3] but they also need to have access to physician time without parental involvement[4]. Physicians' and parents' accounts in our study are aligned with such adolescent wishes as participants were aware of differences in patient preferences. When parents agreed to open communication, they were guided in their supportive actions by teenagers' own behavior and wishes.

Besides parental and physician sources of information, children were reported to acculturate to the hospital setting and therefore knowing more about diagnosis, treatments and illness consequences. Participants mentioned that some children manifested a wish to know more detailed information about their illness and likely outcomes. These patients also went searching online[20,21] or when parents opposed absolute disclosure some teenagers researched their symptoms on the web to arrive at a concrete diagnosis on their own. This scenario exposed children to abundant information (poor prognosis, end-of-life issues and long-term effects) that they may not be able to filter or structure in the same way as when informed by clinicians. Patients may think their situation is direr than it is in reality[22]. This shows how

ineffective restricting child involvement for protection can be: it can trigger patient curiosity and exacerbate fears.

Study limitations

We relied on treating physicians' judgement in approaching parents for interviewing. This may have led to selecting less difficult patient cases or families that physicians had a good relationship with. The study has the strength of capturing both parental and physician perspectives on child involvement and juxtaposing their views to identify challenges to patient participation and risks when restricting involvement. By having the double perspective parent oncologists and in light of reports of limiting involvement, we believe that social desirability tendencies were minimized. Recall bias may have also played a role in interviewees' accounts. Interviewing allowed in depth probing of parental and physician actions and attitudes towards child involvement. This qualitative method may limit to some extent the results' generalizability to other pediatric oncology contexts. However, the issues identified may be relevant for similar crowded oncology treating centers with limited psychosocial services.

Conclusion

Parents' and physicians' accounts paint an image of children's involvement in cancer care that has different facets. It can be described along a continuum: restricted but widening for some patients and then further being enhanced depending on parental and physician behavior. Some parents who adopted restricting techniques, particularly concerning diagnosis disclosure, viewed involvement rather as a practical step or as being unavoidable given the long treatment and different sources of information to which patients were exposed to. Parental explanations further emphasize that child involvement in care is indisputable, despite

the many grey areas in allowing or ensuring patient participation in care. High parental uncertainty and fears related to cancer diagnosis suggest the need for research on how clinicians can support parental communication with patients. Physicians should aid patients in their involvement, separate from actions to soften parents' boundaries. Oncologists and parents should be aware of the hazards of leaving patients with unanswered questions about diagnosis for a long time. Patients may resort to external sources of knowledge and face information overload, no longer being able to pace its rhythm. Professional support is essential in untangling information relevant to patients' situation from worst case scenarios. All physician actions aimed at supporting patients' participation should be paralleled by parental involvement in care.

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'The world is so unpredictable. Things happen suddenly, unexpectedly. We want to feel we are in control of our own existence. In some ways we are, in some ways we're not. We are ruled by the forces of chance and coincidence.'

(Paul Auster)

Chapter II

Decision-making and the inclusion of pediatric patients in care

Decision-making capacity of children and adolescents - suggestions for advancing the concept's implementation in pediatric healthcare

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Abstract

Shared decision-making in pediatric care requires the collaboration of physicians, parents and minor patients. Children's ability to understand and reason about information provided to them regarding disease and treatment is a key determinant of their decision-making capacity (DMC). Physicians are tasked with assessing patients' DMC, but in practice encounter various difficulties related to: (1) inconsistent use of terminology and poor definition of the concept, (2) absence of reliable instruments tested in minor patients, and (3) of a framework of children's and adolescents' developmental abilities applied to DMC. This article explores these three issues in relation to ethical considerations of children's participation in care according to their developing mental and emotional capacities. The results of this analysis refine the concept of DMC in pediatrics and offer advice on its clinical applicability, taking into consideration physician roles and attitudes when assessing minor's capacity. Flawed understanding of DMC in children and adolescents is a serious barrier to achieving adequate participation in healthcare for minor patients. Revisiting the concepts' interpretations and definition and clarifying issues surrounding assessment and child development is necessary for overcoming challenges in this area and improve pediatric practice.

Introduction

In view of the ethical principle of respect for children[1] as persons with developing autonomy and research that points to clinical benefits, minor patients' participation in healthcare is commended by different pediatric associations[1-3]. Shared decision-making, a process in which physicians engage with patients to make decisions is recommended also in pediatrics, thus requiring the collaboration of parents and children[4-6]. The parties' contribution to this process varies depending on the available options and their complexity, and for children and adolescents it is also shaped by their cognitive and emotional development[7]. Children's abilities to understand and form judgments based on the information provided to them define their decision-making capacity (DMC)[4,7] and therefore their aptitude to provide consent for treatment[8]. Valid and informed consent also requires that information is provided in a clear manner and adapted to individual patients' understanding, and that patients express a wish to decide. The essential elements for children's capabilities to consent to treatment - decision-making capacity, information provision and voluntariness - are subjected to physician and parental influences, social customs and regulations, developmental aspects intrinsic to every child, ethical and legal prioritization of best interest or burgeoning autonomy[9-12].

Capacity assessment is the responsibility of physicians and can be carried using the criteria proposed in adults. This includes patient understanding of essential information, analysis of the situation and likely outcomes, ability to reason about and communicate choices[13]. Additionally, in children and adolescents, DMC considerations have to account for patients' evolving abilities in the context of maturation processes and learning experiences[14]. However, in view of children's changing abilities in general and of variations of capacities within a patient when faced with making various decisions that pose different levels of complexities, capacity in minors is not easy to identify. Moreover, assumptions about age-

[1] Patients who have not reached the usual legal age of maturity (18 years old) will be referred to as children, adolescent or minors throughout this article.

based abilities and caution in interpreting capacities by applying stringent standards of reasonableness of choice further complicate DMC assessments[13,15,16]. In fact, the DMC concept in children is poorly understood in practice and inconsistently studied. Most research focuses on separate criteria of capacity assessment, usually understanding, while neglecting reasoning abilities or comprehension of the situation[17].This article explores issues regarding (1) conceptual definitions, variations in terminology and their confusing effect; (2) practical aspects of DMC use and measurement, and (3) the implementation of developmental framework in DMC assessment in pediatrics.

Terminology and jurisdiction

Capacity to make decisions has also legal implications, as it is a pre-requisite of competence. For adult patients, legal competence, understood as the recognition of a person's authority to decide for herself, is presumed. However, it can be overridden on the basis of clinical evaluations of DMC showing flaws in patient understanding of incapacity to reason about a particular task. Children's and adolescents' legal competence is generally not recognized and parents are charged with providing permission for treatment[17]. Nevertheless, minor patients can have clinical DMC and some legislatures have legal provisions that recognize the right of children who display sufficient abilities to make healthcare decisions[18]. Differences in the legal and clinical language used to describe patient capacities to comprehend and balance different information leads to confusion[19,20]. In practice, this can result in broad application of DMC assessments from one medical choice to another without conducting different clinical examinations[13]. For children, differences and dealing with the conflation between the two concepts is essential, particularly in jurisdictions such at the United Kingdom that recognize minors' right to consent to treatment, but ban the power of declining it[19]. Additionally, legislations vary also in whether they enforce age limits when recognizing children's right to

assent or consent to treatment or support participation based on capacities rather than biological considerations. Both approaches have distinct implications for clinicians and patients. Age limits may result in very young children not being viewed as able to have DMC, while adolescents may be presumed as possessing capacity that can be refuted in practice[21]. The absence of age standards has also its own disadvantages, leaving physicians with little guidance and more inclined to appeal to their clinical experience and intuition when choosing to support patients to assume a more prominent role in decision-making.

Operationalization and measurement

Apart from variation in legal norms, physicians also encounter difficulties because there is no reliable tool or framework for examining children's and adolescents' capacities to participate in decision-making[17,22]. DMC has to be assessed for individual patients, taking into considerations personal characteristics when sharing treatment information, and for a particular decision. The standard for DMC should be correlated with the gravity of the outcome for each choice available to patients[23]. At the same time, here are no stringent criteria for how high the DMC should be in relation to serious outcomes[24].

Appelbaum's MacArthur Competence Assessment Tool for treatment (MacCAT-T) investigates all domains related to DMC in adults[13]. However, its applicability in pediatrics is difficult to establish as there is no solid evidence of its reliable use in children and adolescents[13,24]. Efforts to develop a meaningful tool that serves as support for minors' participation in care, sensitive to their developing abilities, needs to include objective parameters of DMC while also taking into account parental judgments[25]. Additionally, as all assessments have to ultimately be judged by physicians, they have to collaborate with minor patients when making decisions in order to build a solid understanding of how the DMC operates in practice[22].

Including developmental factors of DMC in pediatrics

Although DMC establishes which children can participate or make healthcare decisions and which cannot, the participation of all patients in care is unquestionable. As such, those minors who do not have sufficient abilities to meet requirements for DMC upon first assessments should still be involved, as they bear the potential of reaching the threshold for capacity later in the course of illness[11,26]. The developmental aspect is important because although children are not the final decision-makers when they lack DMC, they can have the abilities to understand information and identify a choice. Therefore, physicians have to take into consideration different levels of child participation and recognize that minor patients without DMC can assent to treatment even if decisions are made for them by parents. However, the assent concept itself can be confusing, especially when there is disagreement between child's choices and parents' decisions[27]. In all steps taken to include children and adolescents in care, adults have to be careful not to deceive children and clarify that while it is important for them to also express their preferences, they can be over-ruled by parents[27]. Step by step, repetitive evaluations of child abilities and voluntariness are imperative in ensuring their inclusion in care according to their changing capacities. It assures the adequate transition from being informed, to giving assent and, when meeting DMC criteria, to providing consent[28].

Supporting DMC in pediatric practice

Children's inclusion in care is highly dependent on the attitudes and actions of those involved in their care. DMC assessments are more demanding than in adults and each time require careful individual examinations and reflection on child-specific characteristics and abilities to understand information. This is highly demanding in practice, particularly in view of the issues identified above: (1) conceptual blurriness, (2) lack of instruments, and (3) including the developmental continuum in DMC. Institutional based protocols developed based on

sound ethical and legal principles and with the contribution of pediatricians may open the way for practical implementation of current guidelines on child participation in care[1,3]. Apart from institutional guidance that accounts for different legal provisions, the inclusion of children should be researched in various medical settings and taking into account factors such as disease experience, parenting styles and power-dynamics. Studies should examine the relation between level of participation and adults' attitudes, as well as its effects on all parties involved. Knowledge obtained from both standardized practice and empirical data can then be used to develop a comprehensive framework for assessing the developing DMC in children and adolescents.

Clinicians and researchers should be aware of the perils of implementing standardized tools. In practice, measurements can be arbitrarily applied and be used and interpreted as tests with definite answers[25,29]. In reality, children's behavior is easily shaped by context and relationships. Reliable DMC measurements tools need physician and parent input before and after they are applied with children. Communication with patients and between parents and physicians is important and is one of the elements that can support or inhibit children's abilities necessary for DMC assessments. Moreover, information about individual patient's abilities should be gathered from various sources, including formal and informal care providers[28]. These reiterative processes emphasize the triadic relationships and the shared decision-making specific for the pediatric setting, where decisions for patients require physician, parent and child collaboration[1,15].

Conclusion

Progress regarding children's participation in care and the advancement of clear standards for DMC assessments in pediatrics requires theoretical, empirical and educational work. Research is needed to better understand the development of children's abilities in healthcare

and under specific circumstances, and to build a DMC tool that captures changing and evolving capabilities[13]. Physician training in ethical and legal issues pertaining to minor patients' rights, assent, and principles of respect for persons and best interest is also needed[30]. Ultimately, any objective DMC measurements have to be interpreted by clinicians who need to reflect on those results as they regard an individual patient within a specific context[31]. How to achieve child participation in care should be at the core of analyzing DMC issues and implementation.

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Decision making in pediatric oncology: Views of parents and physicians in two European countries

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Abstract

Background

Decision-making is a highly complex task when providing care for seriously ill children. Physicians, parents and children face many challenges when identifying and selecting from available treatment options.

Methods

This qualitative interview study explored decision-making processes for children with cancer at different stages in their treatment in Switzerland and Romania.

Results

Thematic analysis of interviews conducted with parents and oncologists identified decision making as a heterogeneous process in both countries. Various decisions were made based on availability and reasonableness of care options. In most cases, at the time of diagnosis, parents were confronted with a "choiceless choice" - that is, there was only one viable option (a standard protocol), and physicians took the lead in making decisions significant for health outcomes. Parents' and sometimes children's role increased during treatment when they had to make decisions regarding research participation and aggressive therapy or palliative care. Framing these results within the previously described Decisional Priority in Pediatric Oncology Model (DPM) highlights family's more prominent position when making elective decisions regarding quality-of-life or medical procedures, which had little effect on health outcomes. The interdependency between oncologists, parents and children is always present. Communication, sharing of information, and engaging in discussions about preferences, values, and ultimately care goals should be decision making's foundation.

Conclusions

Patient participation in these processes was reported as sometimes limited, but parents and oncologists should continue to probe patients' abilities and desire to be involved in decision making. Future research should expand the DPM and explore how decisional priority and authority can be shared by oncologists with parents and even patients.

Introduction

Decision making for pediatric patients is no longer a practice solely based on a clinician-centered model[1-4], but features collaboration between three parties[5-7]. Known as shared decision making, this model is the guiding process in all physician-patient-parent interactions in healthcare[8,9]. It requires adequate communication of medical information and options, while engaging all parties to contribute their views and identify values and preferences in order to reach a decision that is best aligned with the patient's best interest, parents' values and rights, and the child's wishes[2,4,6,10,11].

Shared decision making is increasingly complex and challenging when providing care for seriously ill children[5,12-14]. Cancer in particular is a context in which decision making is charged with uncertainty and emotions[15-18]. It is not only a lengthy but also a dynamic process across the illness' trajectory. It often requires striking a fine balance between maintaining hope and sharing responsibility for very difficult decisions[19-22]. Retrospective research investigating parental experience and roles for children suffering from cancer reveal parents' willingness to be included in decisions and their struggle with constraints such as insufficient information, time pressure and poor communication with physicians[19,23]. Parents usually want to share responsibility with physicians for decisions being made. They are less likely to trust physicians and may even experience regret if they perceive their role as marginal[23]. Especially when it comes to making very difficult decisions at the end of life, studies report that physician-parent collaborative decisions provide parents with a sense of having made the right choice[24].

Studies on children's engagement in medical communication and decisions concerning cancer care show that children appreciate receiving information and being involved, though preferences about the degree of participation vary[25,26]. Qualitative studies describe how involvement of children and adolescents in decision making is often limited[26,27]. Due to the

nature of cancer treatment decisions, which often involve to standardized diagnostic, therapeutic, and follow-up protocols, patients do not have a real choice and eliciting preferences is many times futile[27]. Studies have shown that patients may not always be encouraged to ask questions, parents tend to shield children from information they deem difficult, and physicians give children short and technical answers[26,28]. These tactics in turn may lead to exacerbation of children's uncertainty and to children imagining something worse than their actual condition[28,29].

At present there is no standardized blueprint for how the triad should operate and collaborate in making decisions[4,13,30]. This may explain data showing difficulties faced by parents, physicians, and children themselves when engaging in treatment discussions. Given the preference for shared decision making and the difficulties it poses in clinical practice, this study was conducted to explore decision-making processes for children with cancer at different treatment stages and from two European countries. As decision making is influenced by context, we selected countries with different cultural and institutional norms, and diverse legal prescriptions regarding treatment consent for minor patients. While Romanian law supports parental permission and for adolescent patients recommends assent, Swiss regulations stipulate that patients possessing capacity can provide consent, regardless of age[31,32]. However, in both countries a treatment refusal can be overruled based on the child's best interest standard. The aims of this article are to examine the perspectives of parents of children with cancer and the children's physicians on their experiences with participation in decision making. Furthermore, clinician and parental views and attitudes regarding minor patients' participation in discussions regarding treatment choices are also investigated. Child and adolescent wishes and views were sought only in Switzerland and are reported separately[33]. Based on our findings we propose new elements to be factored in when

mapping decision-making processes for children diagnosed with cancer. Study results are therefore framed within the Decisional Priority in Pediatric Oncology Model (DPM)[4].

Theoretical framework: The decisional priority in pediatric oncology model

In discussing treatment options, the DPM considers two characteristics - cure probability and superiority of each option - and distinguishes decisions based on availability of choices that can potentially lead to higher patient benefit. Decisions are classified in three categories based on (1) whether the superiority of a choice is undetermined, (2) whether the two options are fairly equal, or (3) whether a choice requires significant trade-offs between chances of cure and quality of life[4]. Depending on how the two characteristics of any treatment option combine for a given clinical case, the decisional priority can rest with one of the parties involved. That is, the physician or family can take the lead in identifying a preferable choice, assuming priority in decision making. For most decisions, the model recognizes the reality of physician's priority and allocates more space for parents and patients in decisions that have higher uncertainty regarding cure and when choices are similar in relation to expected outcomes. However, authority for approving the implementation of an identified choice (decisional authority) is recognized as belonging to parents, while children's opinions should weigh more heavily as they mature[4].

Methods

This study presents data from participating parents and physicians in Switzerland and Romania, which were collected as part of a larger project investigating views and attitudes on child and adolescent capacity in health care in Switzerland. Interviews with minor patients were conducted only in the Swiss pediatric oncology setting and were previously published[33]. Research involving minor patients, labeled vulnerable by most research guidelines[34,35], was

more challenging in Romania and unsuitable due to the fact that the researchers were in the first stages of establishing a collaboration. The study took place in eight pediatric oncology centers in Switzerland and three pediatric oncology centers in Romania. Four participating centers in Switzerland are university-affiliated pediatric clinics, while the remaining centers are within the cantonal pediatric hospitals. These centers are located in three different linguistic regions (German, French, and Italian) of the country. The three participating centers from Romania are university affiliated and are located in two distinct regions of the country. These centers care for children from all Romanian counties diagnosed with either solid tumors or oncological/hematological illnesses. The study was approved by the responsible ethics committees in Switzerland and the institutional ethics committees of participating centers in Romania.

Participant recruitment

The study sample was a nonrepresentative, purposive sample of volunteer participants of parents/proxies[1] and treating physicians caring for children (8-18 years) suffering from cancer. Participants were eligible for the study if they spoke English, German, French, or Italian (in Switzerland) and Romanian or English (in Romania). Recruitment of proxies was based on physician selection of patient cases that met the study inclusion criteria. Physicians informed the family about the study and asked whether participants would agree to be approached by a member of the research team. Given the sensitivity of the study topic, proxies were informed of the study by collaborating physicians at the earliest 3 weeks after the diagnosis disclosure. In Switzerland, after obtaining proxy permission, the physician would forward their contact details to the research team. In Romania, once the proxies agreed, the physician would introduce them to the researcher in the hospital on the same or following days.

[1] Parent(s) and proxy are used interchangeably in text to mean the adult, legal representative in charge of the minor patient's medical care.

The research team gave proxies detailed presentations about the study and invited them to participate in an interview. If they accepted, a day and time for the interview was agreed upon. For each proxy interviewed for the study, the treating physician's participation was subsequently requested to discuss corresponding patient cases. Interviews with physicians usually took place in their offices, while those with proxies were carried out mostly in a private hospital area.

Sample

Interviews were conducted with 63 participants, 35 (16 physicians and 19 parents) in Switzerland and 28 (10 physicians and 18 proxies) in Romania (Table 1). The total number of physicians participating in this study is lower because five physicians in Switzerland and five in Romania discussed more than one case. Most of our proxy sample included parents, with the exception of three grandmothers in Romania. These grandmothers were involved in patient care from diagnosis onward and were accompanying the patient during hospital stays, and therefore were interviewed as patient proxies. Participants were interviewed only once and the discussions took place either at 3 weeks, several months, or at a maximum of 2 years after the initial diagnosis of cancer in Switzerland and 5 years in Romania. Two parents in Switzerland and one in Romania were interviewed after their child had relapsed or a secondary tumor was diagnosed. In the case of one patient in Romania the tumor was inoperable, and two more proxies were interviewed after being informed that their child developed metastasis. At the time of the interviews patients were still undergoing therapy with a curative goal. Participants' characteristics from Romania were previously described[36].

Table 1 Participant characteristics (N=63)

	Proxies (N=37)			**Physician (N=26)**		
Switzerland	**n=19**			**n=16**		
	Age (years)	**Gender**		**Age* (years)**	**Years of experience in pediatric oncology***	
	44.6 (range 33-52)	Male	n=4	43.6 (range 36-54)	>12	6
					9-12	3
		Female	n=15		5-8	3
					0-4	2
Romania	**n=18**			**n=10**		
	Age*	**Gender**		**Age**	**Years of experience in pediatric oncology**	
	43.3 (range 34-60)	Male	n=1	45.3 (range 30-62)	>12	4
					9-12	1
		Female	n=17		5-8	4
					0-4	1

*Not all participants provided this information.

Data collection

Three members of the research team conducted the interviews (one in Romania; two in Switzerland). Physicians in Romania invited 21 proxies to participate, 3 of whom declined participation due to their child's advanced illness or disinterest in the study's topic. In Switzerland, as parents were selected by physicians and only those interested in the research contacted the study team, the exact number of initial families is not known. Interviews were audio-recorded, except for four in Romania where participants declined and for which detailed notes were taken. Proxy interviews lasted from 25 to 80 minutes and physicians' interviews ranged from 20 to 123 minutes. Interviews followed the same semistructured interview guide that was developed by the researchers based on empirical research conducted on decision making in pediatric oncology by Hinds and colleagues[37]. The guide was divided

into three sections, covering general information about the patient's situation; treatment options; and discussions and decision making and patient's participation in discussions (see appendix).

Data analysis

Tape-recorded interviews were transcribed verbatim in the language in which they were conducted, except for those in Romanian, which were directly translated into English during transcription. Romanian interviews translations were checked by an independent researcher fluent in both languages. Transcript quality was double checked by one member of the research team. Interviews were analyzed using thematic analysis approach[38] and employing MAXQDA 11 software (1995-2015 VERBI GmbH product) for qualitative data. Swiss interviews were transcribed in the original German, French, Italian, or English language and the researchers conducted the analysis on multilingual material. In a first step, three members of the research team open-coded three transcripts to build a coding scheme[38]. For this step, researchers were fluent in all languages of the transcripts that were analyzed in a group of three: English for the Romanian interviews, and French and English for the Swiss. Thereafter, two members fluent in each of the transcript's language coded most of the interviews together using the coding scheme. For the Swiss interviews one pair of researchers fluent in Italian and French and another pair fluent in German and English carried out this step. One researcher was fluent in all four languages and conducted all Swiss data analysis. Throughout the coding process, new codes were added where needed. In a final step, all codes were checked and grouped together under specific topics such as Medical Communication, Decision Making, Inclusion of Children, Lived Experiences, and Hope and Spirituality. All coders came together to discuss the topic of decision making in pediatric oncology to agree upon data interpretation. Thereafter, all coded segments of the 63 interviews related to decision making were sorted out and further categorized. Discussions among researchers led

to the development of three main categories and subcategories. Representative texts for this final analysis step were chosen and where necessary translated from German, French, or Italian to English by two researchers, and were checked by one fluent in all languages. Fictitious child names followed by patient's actual age accompany the quotes.

Results

Our thematic analysis resulted in three main categories that describe how decision making occurred in pediatric oncology: heterogeneous decision making at diagnosis; elective decisions; and decisions outside standard protocol.

Heterogeneous decision making at diagnosis

In both settings, at the time of diagnosis, proxies and physicians described there being little or no room for making a decision. Physicians and their expertise usually played a major role at this time. However, in the course of illness greater parental input and collaboration took place. Parents' and sometimes children's participatory role increased when prognosis was poor or patients were diagnosed with rare cancers with no standard treatment.

Standard protocol as the sole decision

At the time of diagnosis, most parents felt they had no or little choice concerning essential decisions that determine health outcomes. For most cases, the only reasonable treatment option was to follow established treatment protocols for that particular oncological illness (Quote 1 in Table 2). Parents and oncologists reported that if a decision were to be made, then theoretically, it had to be between having and not having treatment. The latter was not considered an option since they were dealing with a life-threatening illness. Likewise, opting for treatment meant following the standard protocol (Quote 2 in Table 2). At the same time and in relation to the option to follow a protocol, for parents the decision to start treatment

was essential, as this had real consequences for likelihood of cure. As one mother put it, it was the "only chance to get to a good result" (Melissa, 10-year-old, Romania).

Parental challenges in considering the treatment protocol

Irrespective of type of cancer, parents and oncologists in both Switzerland and Romania mentioned how treatment decisions, even for standard protocols, bear complex levels of uncertainty. Families were ill situated to interpret all factors that go into determining the best treatment. Parents emphasized difficulties with participating in health care decision making and viewed oncologists' guiding role as paramount. Equally, they appreciated being listened to by physicians when decisions were made (Quotes 3 and 4 in Table 2).

Exercise of therapeutic privilege in decision making

Oncologists' views on decisions considered essential included their primary role in identifying best options. In few centers, physicians reported that at the time of diagnosis they discussed therapeutic choices among colleagues in order to select the protocol to propose to parents. This was regarded as an a priori step in decision making, before involving parents, and was more likely to take place for patients diagnosed with rare tumors. Oncologists described subsequent parental participation as not influential, but necessary in authorizing treatment administration (Quotes 5 and 6 in Table 2).

Table 2 Heterogeneous decision making at diagnosis

Standard protocol as the sole decision		
1	Jake's mother (Patient age 12, Ch)	'There was no, like options. They said, he's at this stage […], he has a high risk group for this, and this is […] the treatment, this is the protocol'
2	Melissa's physician (Patient age 10, Ro)	'In general .... 100% of the cases, parents want to go through with the treatment because they know that this is the only solution. Without the treatment, the consequences are clear.'
Parental challenges in considering the treatment protocol		
3	Desiree's mother (Patient age 11, Ro)	'I don't have the sufficient knowledge to decide ... from a medical point of view what it is best for my child. I understood that there is a protocol, standardized, worldwide and do I personalize it? On what basis?'
4	Charlie's mother (Patient age 14, Ch)	'They [the physicians] took the decisions, they knew, they have the competence, but they listened to us, when I said: […] I have noticed this, they made their considerations, they evaluated things, but they listened.'
Exercise of therapeutic privilege in decision making		
5	Tyler's physician (Patient age 17, Ro)	'Well, when it comes to decisions, I think that the subsequent participation [of the parent] is not significant because essentially when you get the diagnosis, the decision is taken [to follow a specific protocol] … So they [parents] are the ones who make the decision [to authorize the treatment]: "Yes, sir, let us begin".'
6	Zoe's physician (Patient age13, Ch)	'The parents were involved a little bit […] But in the end the parents were not confronted with the questions: Do you want Option 1 or Option 2, but we [physicians] came up with our recommendation, which was developed "halfway-together".'
Shared decision making: poor prognosis at diagnosis		
7	Zoe's physician (Patient age 13, Ch)	'[…] overall it is a rather bad prognosis. And indeed also a really unusual tumor and that was also what bothered the parents […] very much…. […] we also collaborated with professor [specialist

		for that tumor] … and we actually developed the therapy.'
8	Tyler's physician (Patient age 17, Ro)	'Of course, we had a discussion. The parents came in without him [Tyler]. But after that, I had a triadic open discussion with him, together with. […] since this was also a very serious case. And precisely because it is so serious and rare, the treatment has to be extremely harsh. And when it comes to these rare tumors, there is no protocol for them in the world.'

Note. Ro, Romania; Ch, Switzerland.

Shared decision making: poor prognosis at diagnosis

For three patients, physicians described how at diagnosis treatment was completely outside standardized protocols due to the gravity and rarity of the type of tumors in children. Decision making for these cases required specialist collaboration and team efforts to come up with a treatment plan. Under these circumstances parental involvement in decision-making processes heightened as more discussions took place before arriving at a decision (Quotes 7 and 8 in Table 2).

Elective decisions

Participants reported that once treatment was established, parents and children were provided with opportunities to take charge of so-called elective choices that included two categories: those that concerned quality of life and those medical in nature. Elective decisions were distinguished from the protocol decision based on the fact that the health outcome was independent. These optional choices did not have a direct impact on therapy results, unless they meant postponing treatment.

Improving quality of life during treatment

Elective decisions regarding the modality of administering the treatment (orally or through an intravenous line) and adjusting sessions around holidays had the purpose to improve quality of life during treatment. They were made available by physicians as long as they did not interfere or impact the cancer-directed therapy (Quotes 9 and 10 in Table 3). Oncologists also mentioned choices that were in children's hands, such as what devices to use for administering medication. They reported that such involvement of children gave them some control over what was happening (Quotes 11 and 12 in Table 3).

Table 3 Facultative decisions

Improving quality of life during treatment		
9	Jessica's physician[1] (Patient age 16, Ch)	'We try to let them decide where they can, in the sense that, we try to find compromises on how the therapy is implemented: on the checkups, visits, maybe on the nutrition, on going to school, outside activities. But on the other hand, we do no task: do you want to do the treatment or not. If we think that the treatment is worthwhile, we do not leave the choice, we explain why we will do the treatment in the hope they understand.'
10	Dillan's mother (Patient age 14, Ch)	'On his birthday he could stay at home. That he could do. That was an option [we were given] […]. Normally we would have started on Friday, but then we began on Saturday.'
11	Aaron's physicians (Patient age 14, Ro)	'[Aaron asks] what is the treatment? How many days does he have to do it, how are the drugs called and especially when he has to have a puncture done and when he can go home?'
12	Carrie's physician[2] (Patient age 14, Ro)	'"I [Carrie] am afraid of the catheter, physician. Don't put it." I tell her: […] But look, see? You cannot even see your veins anymore. It would be much easier if you had a catheter or a port." And I [physician] say: "Ok, we will think about what treatment you will have after all and we will decide what to do." In the end, there was no need for the port anymore because she will start the metronomic therapy.'
Medical choices		
13	Tyler's mother (Patient age 17, Ro)	'[The physician] said that he [Tyler] should take which one [of the drugs] he wants out of the two because both are good. And then he [Tyler] asked him [the physician] and took [the drug]. So, he [Tyler] got informed about the specific drug. But regarding the cytostatic sessions, it is much more complicated.'
14	Cristiano's mother (Patient age 15, Ch)	'There were two choices: it is a medication that one can, I believe, take four times or ten times. And we opted for the shorter path because Cristiano just wanted to be done. […]… This is a bit more intensive but not the longer treatment course.'
15	Ben's mother (Patient age 14, Ch)	'I was astonished because he started to cry right away and said that he wanted to have children. [and asked] Will I not be able to have any children now?'

16	Angelina's mother (Patient age 18, Ch)	'With regards to that decision (ovum preservation), she really wanted to do the operation... I, as a parent, also listened to the advice of the physicians, what they thought about this. Thus, I understood that the health of my daughter was at risk there. So, I decided that she would not do the operation, that she starts the chemo.'
17	Tyler's physician (Patient age 17, Ro)	'They [parents] might ask especially in the case of girls, but also in the case of boys, especially for the testicular cancer cases. We take out a testicle. But...it happened that they asked questions. […] Unfortunately in our country, the methods of preservation so to speak are still in the initial stages. That is, abroad you can collect sperm, eggs etc. to freeze them even before you start the cytostatic treatment. Here, these types of banks are limited, so to speak, when it comes to money.'

Note. Ro, Romania; Ch, Switzerland.
[1] Jessica's physician also discussed Charlie's diagnosis and treatment.
[2] Carrie's physician also discussed Lisa's and James' diagnosis and treatment.

Medical choices

Some elective choices involved more intricate considerations. With respect to cancer therapy's immediate or late side effects, parents and children were confronted with several medical options. However, these choices, though of a medical nature, posed lower degrees of complexity, and were deemed to be safe if made by parents and patients as they did not interfere with the cancer-directed therapy. One mother mentioned her son experiencing some side effects from the medication prescribed for his heart condition. After consulting with the cardiologist he decided between two drugs (Quotes 13 and 14 in Table 3).

In four Swiss cases, participants discussed the option of undergoing fertility preservation therapy. Parents noted that choices to undergo procedures for fertility preservation weighed a great deal on their adolescent children and three of them clearly expressed the desire to become parents. Despite it being an elective choice, the patients' role was sometimes challenged when parents viewed it as too hasty or likely to delay cancer treatment (Quotes 15 and 16 in Table 3).

In contrast to Switzerland, where fertility options were discussed, one oncologist from Romania mentioned that measures for storing sperm and eggs are usually not addressed with parents and patients. The limited capacity to offer this treatment and the technique's associated costs were considered restrictive (Quote 17 in Table 3).

Decisions outside standard protocol

Some participants were confronted with decisions involving a choice between the standard protocol and a research protocol or clinical trial. For some, such an option was given early after diagnosis, while for others research participation choices arose when standard protocols failed. These decisions differed from those made at the time of diagnosis not because of the point in time when choices were presented, but due to physicians' medical assessments. In

proposing research participation, oncologists either considered the research protocol to be equally effective to the standard one or evaluated a clinical trial to be superior to attempted standard therapies.

Choosing between standard or research protocol

In the Swiss sample, several patients and parents were asked quite early in their treatment to choose between standard and research protocols. Mia's mother (patient age 14, Switzerland) explained, "They [the physicians] said there is either the standard arm or a [research] study on the side. In the end, she [Mia] is in the standard arm". However, she also reported that this was not viewed as a decision to be made by parents (Quote 18 in Table 4). Another parent highlighted the choice made in collaboration with the child to enter a research protocol after the completion of the standard treatment as an additional measure to reduce relapse chances (Quote 19 in Table 4).

Decisions to step out of standard protocol were also made in a few cases as a result of patient's poor response to therapies. In such circumstances, physicians described how they looked for alternative solutions that could increase patient's sensitivity to treatment (Quote 20 in Table 4). In contrast to the Swiss data, parents in Romania did not discuss participation in research in a straightforward manner. The research option was not identified overtly, but some parents did mention children undergoing treatment that was not the standard protocol. Cody Ray's mother (Patient age 10, Romania) mentioned that her child "did not respond to treatment like all the other children" and that the physician had to try something different with her child. However, she did not perceive it as a decision, but as an exercise of physician's expertise (Quote 21 in Table 4).Comparable to parents' reports, physicians in Romania did not discuss proposing as treatment option a research protocol besides the standard one. They only talked about improvements in cancer therapies for children in general. Oncologists in Romania mentioned different standardized protocols based on the

country where they were developed and that they differ in small ways. They further elaborated that the change in the line of therapy took place when the standard treatment did not render the expected results in a patient (Quote 22 in Table 4).

Stepping into the unknown after several treatment decisions

As treatment continued, for 14 patients (7 in Switzerland and 7 in Romania), events arose where additional essential decisions became necessary. Participants in both groups mentioned that these situations often arose in case of treatment failure, relapses, and when standardized options were no longer available. These decisions were made collaboratively with parents and sometimes children (Quotes 23 and 24 in Table 4).

Evident from participants' accounts is also parents' and physicians' unwillingness to let go even when the illness took a downward turn where patient's demise could be anticipated. Thus, both parents and oncologists felt that all should be done to make sure that patients got all chances to fight against their odds (Quote 25 in Table 4). Typically, oncologists believed that in these complex situations the decision had to be more up to the parents and patients. However, the grim circumstances and the unlikely cure were not openly discussed with patients (Quotes 26 and 27 in Table 4).

Table 4 Decisions outside standard protocol

Choosing between standard or research protocol		
18	Mia's mother (Patient age 14, Ch)	'But speaking about making treatment decisions, I would say, in the end, we as parents, did not really make a decision, the treatment is already there, that's it'
19	Sam's father (Patient age 10, Ch)	'After the first nine cycles [of chemo] plus radiotherapy plus surgery, we could decide whether we wanted half a year of oral cytostatic drugs [as part of a research protocol]. […] During the first part [of the treatment], he [Sam] and we, already decided that we wished to take part in this study.
20	Angelina's physician (Patient age 18, Ch)	'It was a [diagnosis] without metastasis, and with a relatively good prognosis but dependent also on the tumor response. And she did not respond well, she then belonged to the poor responders. Then we discussed with her [that] there is this choice to treat her according to the standard arm [of protocol] or to use a chemotherapy that is part of a study, with the aim to intensify [the chemo]. We made the decision together with her to treat using the standard therapy.'
21	Cody Ray's mother (Patient age 10, Ro)	'So for around two weeks, even with cytostatics, he did not respond to the treatment. Finally yes, but the risk of relapse is very high (s*obbing*). […] I do not know too much. So how the treatment will be... So...My understanding is that the next treatment will be, so it will not be like the one [standard protocol] for the others.'
22	Tyler's physician (Patient age 17, Ro)	'So there are [different] therapeutic groups... […]So there are protocols of phase 4, meaning well established. And there are those of other phases that are less well proven (evidence-based). Of phase 3, 2, and 1 (phase 1 clinical trials), which are experimental. So the problems arise when you don't have a 4 phase protocol that is well established and that you know that leads to results between certain percentages and that has well proven results on thousands of children already.'
Stepping into the unknown after several treatment decisions		
23	Celine's physician (Patient age 13, Ro)	'What happened is that the second time she [Celine] had a relapse, she [the mother] refused a new surgery, claiming that the surgery risks are too high. […] But she said, that she [Celine] has to have the chemotherapy and radiotherapy. Ok, I told her that we do not have that many chances if the surgeon does not take it out from there. But, in the end, she [the mother] agreed to the treatment

		[chemotherapy without a second brain surgery].'
24	Cristiano's physician (Patient age 15, Ch)	'[After the second relapse] then it is a very experimental treatment. […]Well we were not going to give up. Another option would have been that we do palliation, gentle chemotherapy, as long as possible to hold a good quality-of-life. That was something that we [as physicians] would have had to accept also … But it was clear for the parents: we are going to try this possibility [experimental treatment]. I think, the parents talked with him about it [also].'
25	Christiano's mother (Patient age 15, Ch)	'[He asked] "Mum, what was the third [option] that he explained to us? I did not quite understand that." And then I said: Well, I don't know how to explain that to you… And then he said: "Right? I did get that right, they would just let me die; and I don't want to die yet…" And he clearly told the physicians and they […] said: "Yes, I think we are dealing with a fighter".'
26	Tyler's physician (Patient age 17, Ro)	'So the problems arise when you have exhausted the [standard] protocol or he [the patient] had a relapse and in that moment you have to use the second or third line (therapies). From that moment onwards you step into… problems because the second and third lines are not proven so clearly. There are more options and in that moment... indeed, in that moment the parents` decision regarding those proposals is more significant, no?'
27	Serena's physician (Patient age 14, Ch)	'Well, we evaluated if we could operate again. […] But the neuro-surgeons said that they did not want to operate because that would cause more harm. And one option, of course, would have been not to do anything … But that was out of question for everyone. […] She (patient) was present. But I think I presented the option not to do anything when the patient was not present.'

Note. Ro, Romania; Ch, Switzerland.

Discussion

Results from this study highlight decision making's complex and heterogeneous nature in pediatrics[5,9,14,15]. At the time of diagnosis, physicians' guiding role was dominant as they identified the appropriate treatment and a suitable protocol for the particular cancer and advised starting the treatment. However, in the case of rare malignancies, decisions outside standard protocols were made that required not only consultation with different specialists, but also more discussions with families, highlighting their participatory role in decision making. Additionally, for elective decisions, many times parents and patients had a prominent role. Therefore, there was a shift in control from physician to family when choices affected quality of life without jeopardizing treatment outcome. The heterogeneity of decision-making processes is illustrated by the choices offered to some families to opt for research protocols when prognosis was poor.

Our research shows that the central focus of decision making in both countries and in reference to essential and elective choices was the child's best interest. Parents and sometimes patients made various choices regarding treatment, medical tests, or care procedures. They distinguished decisions based on the number of options offered by physicians - only standard protocol or opting between research and standard treatment - and based on their content and significance for health outcomes - essential decisions versus elective ones. These constitute different decisional levels, at which decisional priority is assumed by one party or shared, depending on context and available choices. These levels can be incorporated into Whitney's two-dimensional decision plane[4], which also describes decision making as heterogeneous. Based on the study's results, we explore how the model fits the reality of clinical practice and challenge decisional priority and authority roles attributed to parents and/or physicians. A standardized blueprint for triadic decision making has to be more sensitive to switches in decisional priority and authority among parties. In

decision making for pediatric oncology patients this must also encompass more strengthening of patient involvement in this process.

At diagnosis, for most patients decisions were perceived as a "choiceless choice"[39] situation for parents and children. Oncologists proposed a standard protocol because, in their view, it was the best treatment available for the specific cancer diagnosis. Interviewees described these decisions as essential insofar as they had a direct impact on chances of cure. Coyne and colleagues[27] describe them as "no real decision". This raises questions because parental authorization of treatment is still necessary even when decisional priority lies with oncologists[4]. In considering physicians' leading role in treatment selection at diagnosis, it should be emphasized that families still participate in voluntary decision making. What appears a "choiceless choice" for families is not the result of physician constraint or preexclusion of other similarly good options, but the consequence of a constraining context[40]. In pediatric oncology, decision making is an ongoing process and participants identified several instances along their children's illness when they were faced with one or more essential decisions. As such, reflections on a model of decisional priority and authority should not obscure parties' interdependency, particularly that of parents and oncologists. Our results reflect this potential conflict: Parents discussed their challenges with authorizing decisions and simultaneously described physicians' role as inextricable to parental permission for treatment. Parents expressed how they trusted physicians and how medical expertise was crucial in making treatment decisions. This is consistent with previous research reflecting the influences of social factors on decision making[27,39], which is subjected not only to determinations of risk and benefits, but to a broader social interaction context in which relationships and trust factor in[39]. This expands the notion of physicians' decisional priority in relation to chances of cure at the time of diagnosis as framed by the DPM. Whitney's model acknowledges the pivotal role of oncologists in prioritizing the treatment option based

on medical assessments[4] when, in fact, parents invest and support oncologists with decisional priority through their trust and appreciation of their professional recommendations.

Some parents and patients were confronted with making essential decisions when they had to opt between standard and research protocols. This finding complements research on how, in pediatric oncology, parents and children could realistically make only small decisions[27]. Additionally, for other patients for whom therapy failed, decisions had to be made in consideration of how invasive or aggressive the treatment should be. Choices included experimental treatment and palliation with low doses of chemotherapy. These decisions were subsequently characterized by increased family involvement, especially in view of the lack of choice at diagnosis. Parents and sometimes children had greater input in late phases of the illness. This greater sharing of decisions in the context of relapse or terminal illness is observed in studies investigating end-of-life decision making and transition to palliative care[24,37,41]. The DPM[4] includes such essential decisions, termed "no best option". These decisions depend on context and must be shared between families and physician or may be fully assumed by either parents or patient. These elements proportionately define the benefit margin to be expected to result from choosing one option over the other, as the medical superiority of the available treatment options is not clear[4].

Families had opportunities to make elective decisions, similar to nursing or care procedures identified in other research[27]. They were confronted with several decisions involving at least two options around how and when care will be delivered. These decisions mainly consisted of elective choices, usually whether to take drugs orally or intravenously, that paralleled the more complex cancer-related treatment. Participants described how patients were encouraged to make such decisions or their preferences were accommodated as much as possible. The leeway parents and particularly children had to opt for something was greater because the final decision would not interfere with cancer therapy. In Swiss interviews, fertility

preservation was one of the available elective decisions. These decisions were significant and of higher complexity than daily care options. However, patients' role was overshadowed in some cases when final decisions were more a result of parental persuasion. Adolescent preferences can be swayed by parents' and oncologists' prioritizing the start of treatment over desires to undergo fertility preservation. Young females may receive disparate support for fertility preservation decision making compared to males. This can partially be explained by the relatively uncomplicated technique to collect and preserve sperm as opposed to egg and ovarian tissue[42,43]. These elective options, especially complex ones, and the decisional priority clash that may emerge are not explored by Whitney's model[4], which overlooks that in the course of illness parents and patients are likely to face multiple decisions outside cancer-directed therapy. These options may be important in giving patients back some of the sense of control that usually is lost in the context of a cancer diagnosis[25,27]. The DPM also does not address the more difficult decisions around opting for medical procedures that are highly personal (fertility preservation) and can impact health outcomes, or who should give authorization for them.

Regarding opportunities for treatment decisions, we observed some differences between countries. Fertility preservation was an elective choice discussed only in our Swiss data, which may be explained by the existence of a Swiss therapeutic network since 2010[44]. In Romania, fertility preservation is available, but offered only in private hospitals and practices[45], and, as mentioned in Swiss interviews, physicians and parents may discourage such treatments when they cause delays to cancer therapy. Additionally, the procedures for preserving fertility are expensive, which would restrict access for Romanian patients, but also poses a barrier in Switzerland, where oncofertility is not covered by health insurances[44]. In Swiss interviews both oncologists and parents mentioned choices regarding research participation, whereas in Romania participants did not openly address such options. The latter

appear to have fewer opportunities to make or participate in essential decisions. This finding has a twofold consequence for the DPM. First, the threshold for requiring patient assent may be higher in research than clinical care - and patient dissent is ethically binding[8]. Therefore, for these decisions authority may be equally shared between parents and children, who can even make the final decision, provided they are mature enough[46]. One of DPM' pillars refers to the decisional priority and authority necessary to reach a decision and which is assigned to oncologists, parents, and sometimes patients[4]. While the priority in selecting an option is presumably shifting between parties, the DPM fails to explore whether the authority to decide truly lies with parents only or should be shared with oncologists and even with children[30]. Regarding research participation, differences between Swiss and Romanian legislation about recognizing patient assent significantly influence the sharing of decisional authority with patients. Under Romanian legislation, children's interests are promoted by parents[31] and, similar to the United States, it is only when parents are neglectful that the patient's wish can weigh more heavily in medical decision making[47]. Swiss law supports the self-determination of competent patients, independent of age[32], and therefore promotes minors' participation and decisional authority when it comes to research also based on the requirement of voluntariness.

Second, research suggests that decision making may be influenced by factors such as institutional organization and differences in clinical communication[46,48,49]. In our study, it appears that families in Switzerland were explicitly offered opportunities to participate in essential decisions concerning research. This difference may partially be the result of varied practices of informing patients about and offering them clinical trials[48], of seeking permission or assent and supporting patients and families in making decisions[46,50]. It may also be the result of a conflation of protocols for standard and experimental therapy, as many standard protocols have some research component[51,52]. The research topic was more predominant in

Swiss interviews, as only some physicians in Romania mentioned research trials in general, while parents rarely talked about different or new protocols. This country difference may reflect research capacities and infrastructure imbalances within Europe and disparities in access to the newest clinical trials[53]. It could also stem from lack of protocols for communicating with families about research, limited capacities in terms of multidisciplinary support (such as a research nurse, ancillary personnel), and physician lack of time due to high number of patients[53-55]. These issues can be observed even in countries that have well-developed health care systems, but are more likely to affect smaller, regional clinics or crowded hospitals in both countries. However, decreased research infrastructure and increased patient burden per physician are typical of Romanian hospitals. Such contextual issues are not fully explored within the DPM, thus limiting its usefulness for diverse clinical situations or settings.

Furthermore, the model fails to recognize external influences, such as consultation with other family members to identify choices, which shape decision making and go beyond evaluations of medical facts[39]. It overlooks potential for disagreements or open conflicts not only between physicians and parents, but parents and patients[30]. As our data suggest, lack of consensus may occur even when deciding on elective choices and it is more likely that patient's voice will dim when it does not match parents' and physicians' views[47]. As such, it seems intuitive that in a theoretical decision-making model, patient role should be considered independently of parents' and not merely collapsed under that of the family[30].

Study limitations

This study's results need to be interpreted in view of its strengths and limitations. Data analysis was conducted on multilingual materials (English, French, German, and Italian), which may be challenging for interpretation. However, all coders were fluent in English and

the Swiss data coders were always grouped in language pairs according to fluency, with one researcher proficient in four languages conducting all stages of analysis. Additionally, all other researchers in the team have very good knowledge in all languages. They participated in discussions on data and conducted the final analysis for the decision-making topic. Interview participants are from two distinct European countries with different health care systems, disparities in infrastructure, and care services provided within pediatric cancer units[53]. Despite these differences, participants in both countries described decision-making processes in similar ways, and patterns across the data overlapped to a great extent. Additionally, the study captures both parental and oncologist perspectives and therefore offers a comprehensive representation of the communication and experiences of making decisions regarding cancer for minor patients. Perhaps in view of some parents' refusal to participate due to their child's being at the end of life, the results are more representative of decision making for patients for whom treatment has a curative goal, including cases of poor prognosis. We did not include patients' voices, which might have given a clearer and perhaps more comprehensive portrayal of decision making. However, interviews conducted with child and adolescent patients in Switzerland, published elsewhere[33], support the decision-making processes described in this study. Parents and physicians did mention instances of patient participation in expressing preferences, choosing options, or making elective decisions. The results may be biased by the fact that we interviewed participants about decisions and experiences that in some cases took place as long as 12 months or more in the past. Additionally, participant inclusion may be biased by personal views and interest in interview topics. Social desirability may have also played a role in the way participants described certain situations. Innate to the design of qualitative methodology, results are not generalizable to all pediatric oncology settings. Nevertheless, the diversity of pediatric oncology units included in this study, from university-affiliated hospitals to smaller clinics

belonging to two European countries with different health care systems, is likely to be representative of oncology care in pediatrics in both high- and medium-income countries. Results also represent a deep exploration of experiences around decision making in these two settings.

Conclusion

Overall, results from our study show that decision making in pediatric oncology evolves along a continuum, with families and physicians having to sometimes make decisions at diagnosis based on a sole choice, only to be presented later in the illness' course with other decisions involving more options and of different complexity. These processes match Whitney's DPM[4] and point to its adequacy in guiding clinical practice. However, in moving forward to empirically support a decision-making framework in pediatric oncology, researchers need to consider its flexibility to accommodate contextual differences and potential cultural nuances. Clinicians, ethicists, and researchers should broaden the concept of collaboration to bring the focus not on who makes the decision, but how a decision is being made. Elements such as the distinct role of patients, parents, and oncologists, diverging opinions and conflicts, and the existence of different types of decisions have to be incorporated. Taking into account the different constraining factors - no choice or difficulties in distinguishing between several choices, uncertainty, and time pressure - communication and sharing of information among the three parties seem to be the optimal manner of increasing both parental and patient participation in these processes. The DPM is a good starting point and reflects the constraining context of a cancer diagnosis, but fails to recognize the many ways oncologists can share decisional priority and/or authority with parents and even patients. Future endeavors to improve decision-making frameworks should place more emphasis on physician-parent-patient collaboration and the support families need

to enhance their role in these processes. Additionally, the importance of communication in reaching decisions with which all parties can comply and adhere to needs more acknowledgement.

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Appendix. Examples of questions from the semistructured interview guide for parents and physicians

Parent Interview Guide

General information about the patient's situation

Let me start by asking you about your family. Could you tell me about them?

Please briefly describe the circumstances regarding your child's diagnosis?

Was your child present during the diagnosis discussion? Why or why not?

How did you feel about your child's presence in regards to the diagnosis communication?

Since the diagnosis was made, have you informed your child about the diagnosis? How did you do so? Could you tell me the words you used? For example: what was the first sentence you used to start the discussion?

Treatment options, discussions, and decision making

What were the treatment options?

Who was present when treatment options were discussed?

How were these treatment options described to you and/or your child by your physician?

If your child was not present during treatment discussions, have you discussed treatment or non-treatment options with your child?

Patient participation in discussions

In your previous response, you stated that your child was present (absent) during treatment discussions. Could you explain the reasons for this?

Why do you think it was necessary/appropriate to include (exclude) your child from these treatment decisions?

Under what conditions would you absolutely include (exclude) your child in such discussions? Please explain these conditions.

Physician Interview Guide

General information about the patient's situation

How long have you known the patient and his or her family?

Who did you disclose the diagnosis and prognosis to? If your patient was not present, please indicate why.

What was the parent's reaction when the diagnosis and prognosis were made? If you discussed the diagnosis/ prognosis with the child, what was the child's reaction?

Who initiated this discussion related to diagnosis and prognosis with the parents and/or the

child? How did you feel during the discussion?

Treatment options, discussions, and decision making

What were the treatment options?

Who did you discuss the treatment options with?

How did you describe the treatment options to the child and/or the parents?

How comfortable did you feel discussing treatment options? Please explain.

Patient participation in discussions

In your previous response, you stated that the child, i.e., patient, was present (absent) during treatment discussions. Could you explain the reasons for this?

Why do you think it was necessary/appropriate to include (exclude) the patient from these treatment decisions?

Under what conditions would you absolutely include (exclude) patients in such discussions? Please explain these conditions.

Generally, when parents include (exclude) their child from making or taking part in such decisions, what kind of reasons, for example values and attitudes, do they give you?

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Putting patient participation into practice in pediatrics - Results from a qualitative study in pediatric oncology

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Abstract

Insufficient recommendations and practical steps make the implementation of the ethical desirability of child participation in care challenging. A qualitative research study conducted with 17 minor patients, 19 parents and 16 oncologists explored the practical experiences of these participants with child involvement. Results show that for children and adolescents with cancer participation is defined by three themes: modes of participation, regulating participation and influencing factors. In pediatric oncology the involvement of patients in care is complex and the subject of many challenges for physicians. Ultimately, clinicians are best positioned to positively shape parental influences on involvement and ensure space for children's expression of choices.

Introduction

Adequate participation of children and adolescents in care is ethically motivated by promoting respect for their developing autonomy[1,2]. In practice, involvement can also facilitate understanding about the illness and mediate psychological wellbeing[3,4]. However, participation of minor patients in care is difficult to achieve because it is individual based and has to be calibrated to personal abilities that are evolving in the course of treatment[5,6]. This requires close physician-parent communication and sharing rather than exchange of information between all parties[3,7]. Physician training may be inadequate to meet all these standards[8]. In view of these issues, the present study aims to examine how participation is materialized in pediatric oncology centers in Switzerland.

Methods

Semi-structured interviews were carried separately with parents, patients and treating oncologists recruited from 8 centers of the Swiss Pediatric Oncology Group. Eligibility criteria included: patient age between 9 and 17 years and receiving treatment at any of the centers. Ethics approval was obtained from Ethics Committees from each of the states (cantons) where the centers were located. All participants offered informed consent before the start of the interviews. Data was recorded and transcribed verbatim. Analysis was carried by using MAXQDA software and employing thematic exploration of interview themes.

Results

Fifty-two interviews were conducted with 19 parents, 17 patients and 16 physicians, with five oncologists discussing two patients each. Participant accounts portray described child and adolescent participation in relation to three themes: (1) mode, (2) regulating participation and (3) influences.

The first theme describes the different ways in which children were involved in their care. Some patients were present from the first parent-physician discussions and received unfiltered information. Others were informed by parents after physician-parent communication took place. Most children would switch between these two modes of communication throughout treatment. Besides participating in discussions, children were reported to actively engage through asking questions, gather knowledge by observing and sometimes express preferences for care procedures that accompany treatment. This step resulted in the inclusion of children in some of the decision-making processes related to care.

Children's involvement in care was subjected to actions to regulate their participation. These were exerted by patients, parents and physicians. Parental strategies concerned limiting participation by controlling the information shared with patients. Together with physicians, parents also used pacing of information as a means to avoid overwhelming children. Children also used the pacing mechanisms to allow themselves to get used to difficult information.

In general, participation was shaped by parenting styles, adherence to treatment and following strict protocols that inhibit significant participation in treatment decision-making and by a desire to foster compliance in children.

Discussion

Data from this study highlights practical ways in which children and adolescents are participating in care during cancer treatment. The inclusion of patients is usually achieved by sharing information and inviting their opinions in decision-making processes. However, there are limits to their participation that are controlled by parents and physicians. These are evident in what concerns treatment decisions or increased risks due to poor prognosis[7]. Similarly, children and adolescents use mechanisms of pacing information and observation to establish the level of communication and participation that best fits their needs. Involvement

preferences differ among patients and can change in the course of illness for individual children. Physicians duty to engage with children is therefore very complex and made more difficult by parents' wishes and views on how best to involve their children in care[5,8]. Adequate physician training on communication with families and children and on how to reduce the impact that cancer experiences have on them is necessary[9].

Conclusion

Professional guidance is insufficient when dealing with patient participation in pediatric oncology practice. There are several non-static ways in which patients are involved in healthcare. Moreover, patients, parents and physicians have their own mechanisms with regulating participation and dealing with factors limiting information sharing. Physician specific training on how to adequately identify patient wishes and support them in view of parents' influences can improve children's modes of participation.

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Parents' and patients' experiences with pediatric oncology care in Switzerland - Satisfaction and some hurdles

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Abstract

Children diagnosed with cancer and their families experience significant distress during treatment. Ensuring their satisfaction with healthcare professionals' support is important for improving health outcomes and delivering patient cantered care. This study examines parents' and patients' satisfaction with care in Swiss paediatric oncology units and identifies areas of improvement. Qualitative interviews were conducted with parents and their children undergoing treatment for cancer. Data was subjected to thematic analysis to explore the topic of satisfaction with care. Thirty-two participants were interviewed about their experiences with care and clinicians. Satisfaction was rated high by parents and children who identified good communication, clinicians being available and approachable, making extra efforts to support families based on individual needs as improving their experiences. Suggestions on how to improve the quality of hospitalizations referred to reducing fragmented care, improving communication among healthcare workers and between departments, providing solutions to language barriers, and reviewing approaches when offering reproductive health choices. Working towards addressing the issues highlighted by participants can have reverberating positive effects on improving patients' and families' experiences, but also medical care and communication within the organization.

Introduction

Clinician behavior and communication when caring for patients diagnosed with cancer and their families contribute to improving their experience with the illness[1]. Physicians' ability to relate to children and their parents influences the building of trusting relationships and higher satisfaction[2]. Apart from clinicians' characteristics and social capacities, families appreciate access to services and competent care, as well as adequate information provision[3,4]. Research on satisfaction in pediatric oncology has mainly focused on end-of-life care and studies do not include the patient perspective[5,6]. We conducted a study in Swiss pediatric oncology units to examine parent and child views and satisfaction on provision of care, as well as the issues they encountered.

Methods

Interviews based on a semi-structured guide were used to collect data from 8 pediatric oncology centers in Switzerland. Participants were parents of children receiving treatment for cancer at those centers, as well as patients between 9 and 17 years old. Data was analyzed through repetitive readings and initial open-coding of all data sets. Text excerpts that relate to the topic of satisfaction with care were then selected and subjected to thematic analysis.

Results

Interviews were conducted with 19 parents, 15 mothers and 4 fathers, and 17 children and adolescents. Most families were in treatment for less than one year. Participants expressed several positive aspects during the care they received, most related to clinicians' interpersonal skills. Parents and patients identified as valuable the caring of the staff, good communication and taking extra steps. Information provision and sharing among physicians, parents and patients was particularly tied to higher satisfaction. The manner in which physicians

approached parents and patients by communicating clearly and not in a condescending way, gave families a feeling of being supported.

Participants reported some difficulties related to transfers to other units, receiving treatment in a different center and break-down in communication between different physicians. These gave parents additional reason to worry and feel overwhelmed. Additionally, communication that was not tailored to their needs lead to dissatisfaction and in the case of one patient, it was made even more serious given language barriers. Discussions on preserving fertility in teenagers also left parents feeling unease and confused when left to talk about it with their child. Children's reports on care provision gravitated around practical aspects to make them more comfortable while being in the hospital: difficulty to handle multiple tests and procedures in one day, the sterile environment and limited selection of entertainment games.

Discussion

Results from this study offer a varied perspective on aspects of care and provision of services that influence patient and parent satisfaction, and contribute to wellbeing. These views may be easily overlooked during crowded round or routine visits. Considering the many stressors that affect families undergoing cancer treatment, clinicians should address the concerns raised by participants in this study.

Communication, when carried in a sensitive way, offering clear information and responding to individual needs, was the central factor to influence parental and child satisfaction. Similarly, difficulties in communication led to dissatisfaction and restricted parents' participation in their child's care or their understanding regarding treatment and procedures[7]. Balancing individual parents' information needs is not an easy task and physicians should receive additional training support on how to deal with it[8]. Parents perceived discussing fertility issues and informing children as a very delicate issue. Therefore, good

communication may require physicians to pay additional efforts when proposing preservation therapies and offer better parental support[9]. Additionally, communication improvements are warranted also within the medical team and attention should be paid to difficulties arising from discontinuation of care.

Language problems were identified by one parent, but given the diversity of Swiss population and the existence of 4 national languages, this is unlikely to be an isolated hurdle. This issue in particular raises the risk not only to increase parent anxiety and result in dissatisfaction, but to limit parental comprehension on treatment and important aspects of care. Developing protocols and structures to offer translation services for such patients and their families should be a priority.

Conclusion

Parents' experiences with pediatric oncology care highlight a general satisfaction with clinician competencies, both medical and personal. However, most significant difficulties detailed in this study remain in the area of communication.

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'Abby is dying. She is eligible for the drug study, but it holds only the most remote prospect of helping her. At the same time, the drug may be risky and could accelerate her death. (…) Abby looks at me and asks whether I think she should be in the study. (…) Do I owe it to all the children who will get cancer in the next several decades to say yes (to her participation) or should I protect Abby from further suffering and say no?'

(Eric Kodish, MD; 2005[1])

Chapter III

Ethical reflections on clinical research involving children

[1]Kodish E. *Ethics and research with children. A case-based approach*. New York, US: Oxford University Press; 2005.

The Vulnerability of the Individual Benefit Argument

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Abstract

Vulnerability is a highly debated topic in research with human subjects. In "Shared Vulnerabilities in Research" the author analyses the adequacy of the U.S. Code of Federal Regulations 45 (CFR 45) to protect vulnerable human research subjects. He concludes that research conferring no direct benefit conducted with a subject from a vulnerable group cannot be morally justified on the grounds that the beneficiaries of that research share the subjects' vulnerability. While we agree that the paper's central argument is not only compelling but also valid, we suspect that the argument is unsound, as it rests on the faulty premise that one (A) suffers for the benefit of another (B). The argument thus focuses on the individual, rather than addressing the collective (group level), as stated by the regulation's broad wording. Hence, the author wrongly extracts A from his group (of vulnerable subjects). Consequently, any opportunities or benefits that may come to the group will exclude A. We determine that it does make a moral difference whether the beneficiaries share the research subjects' vulnerability.

Vulnerability is a highly debated topic in research with human subjects. Although the construct of vulnerability is widely used, there is no consensus on its definition or on the practical application of vulnerability to protection of research participants, e.g.[1,2]. In "Shared Vulnerabilities in Research" Chwang[3] examines whether the U.S. Code of Federal Regulations, title 45, part 46 (CFR)[4], offers adequate protection to vulnerable human research subjects. He concludes that research conferring no direct benefit conducted with a vulnerable subject cannot be morally justified on the grounds that the beneficiaries of that research share the subjects' vulnerability[3].

While we agree that the article's central argument is not only compelling but also valid, we suspect that the argument is unsound, as it rests on the faulty premise that one (A) suffers for the benefit of another or others (B). The article's central argument thus focuses on individual benefits to B, rather than addressing the group of beneficiaries as a whole, as implied by the regulation's broad wording[4]. Only by doing so can the author formulate the assumptions to support his thesis "that harmful research with to-kind benefits is no less wrong than harmful research with not-to-kind benefits"[3].

The subtle difference between individual and group benefits is also considered in the Belmont Report[5] when fairness is evoked as a guiding principle in research subject selection. In discussing the nature and scope of risks and benefits, the Report points out that these "affect … society at large (or special groups of subjects in society)"[5]. Therefore, it is concluded that while the protection of subjects should always prevail, group interests such as expected benefits in the form of generalizable knowledge may sometimes be sufficient to justify the risks of research[5]. The group argument is compatible with the view that ethically conducted and scientifically relevant research that is likely to yield generalizable knowledge constitutes a social benefit[6]. In research, the social benefit is understood as "something of positive value related to [the] health or welfare"[5] of a particular group[5,7,8]. Likewise, group theories state that

the group as a whole would derive some gains if the group's objective (i.e. generalizable knowledge) is achieved[9]. In this respect, the thesis should not be framed solely around the moral difference of "shared vulnerabilities" between individuals A and B, but also on another key difference: whether individual A is a member of the particular group that benefits.

Indeed, we assume that any research subject A, who is vulnerable and shares that vulnerability with others, is part of a group together with these others and not with any others. As exemplified by the CFR[4], this particular group could be one of several classes: pregnant women, fetuses, and neonates; prisoners; or children. By accepting the label of vulnerability typified by the CFR, the author himself tacitly admits that the vulnerable research subject A is part of a group of vulnerable subjects[3]. This does not equate to saying that A will receive direct, indirect, or ancillary individual benefit from research participation, while being part of the group. Rather, it seems more plausible that any opportunities or benefits that flow to the group as a result of A's role as a research participant would naturally flow to A, too, as they will to the group as a whole. In fact, all individuals in the group will not only seek their individual benefit, but also a group benefit that equates to the social benefit of generalizable knowledge. As in group theory, individuals' action in research with no direct benefit is driven by necessity (the knowledge cannot be gained in any other way) and is dependent on individual behaviors (some vulnerable individuals might consent, while others might not)[9]. Realistically, of course, there will be cases in which research does not yield benefit to the targeted vulnerable group as a whole, but these facts cannot be established before the research is conducted. In such cases, the priority of the research question and of the expected social benefit, as well as the scientific relevance and rigor of the research, are essential to its ethical justification.

Although, "there is no settled framework for how potential social benefit should be balanced

against individual risk"[10], the group benefit should not be excluded from any such assessments and should be explicitly disclosed to individual research subjects[11]. Ultimately, it does make a moral difference whether the beneficiaries share the research subjects' vulnerability, because the shared vulnerability is the core reason around which the group that benefits from research with vulnerable subjects is formed.

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Body matters: rethinking the ethical acceptability of non-beneficial clinical research with children

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Abstract

Children's participation in research that does not provide a direct benefit is necessary for the advancement of treatment and improving health outcomes in pediatric populations. However, the ethical discourse is split between pro-research arguments and cautionary discussions on risks and potential exploitation of some children for the benefit of future minor patients. This article addresses this controversy in relation to Wendler's thesis of fostering altruism and by taking into account risk-benefit assessments. It further adds a phenomenological approach to conceptualize children's bodies and their vulnerability in research not as mere objects of research, but as means to contribute as persons to a project that may not offer any clinical advantage to them. Therefore, we propose moving away from viewing children in non-beneficial research as body-objects and acknowledge their contribution as body-subjects, independent of their ability to consent.

Introduction

Research that does not offer the potential of clinical benefit for participants is subjected to strict ethical criteria and justifiable in adults by the doctrine of informed consent[1-3]. Justifying the conduct of non-beneficial research with children is more complex. Legally they are incompetent, therefore incapable of giving valid informed consent and ethically they are labeled vulnerable, deserving additional protections[2,3]. Different solutions to allow research with no direct benefit to child subjects have been formulated but did not result in finding an ethical consensus: requirement for substituted consent from a legal representative, enrollment only for research that has high social value, that imposes minimal risk for children and cannot otherwise be conducted, and the assent of minor participants[4]. While some scholars argue that a degree of exploitation is unavoidable, but acceptable as long as children's fundamental rights are safeguarded[5,6], Ramsey declares that research with no prospect of direct benefit to the child participant is always unethical[7]. Studies that do not seek to improve or cure the illness of the individual child enrolled are morally wrong because parental consent cannot repair the harm inflicted by bodily invasions[7,8]. This view is too restrictive and risks exposing children to harmful experimental treatment without the chance of gathering solid knowledge to advance pediatric therapies[9]. This article contributes to the debate by discussing a phenomenological view on the permissibility of enrolling children in non-beneficial studies. We leave aside arguments around risks and benefits and analyze the morality of enrolling children by looking at the relationship between children as persons and their bodies to avoid objectification. This analysis expands Wendler's thesis of "contribution to a valuable project" to incorporate the body-subject view of children's offerings to knowledge independent of their presumed incapacity to consent.

Protection and access

Research restrictions and ethical dilemmas concerning non-therapeutic research with children are the result of past abuses and mistreatment[10]. Using some minor patients for the purposes of developing treatments to benefit others may be acceptable on consequentialist grounds, but it remains morally wrong as it instrumentalises some children[11]. McCormick pointed out that even under these circumstances, children can derive a moral benefit by acting as agents belonging to a community they can contribute to[12]. Guidelines incorporated this view of children as social beings who have an indirect benefit and allowed their participation in research without direct clinical benefits on a "last resort" basis – after tests were done in animals and then adults[8,13]. This approach was too narrow to help advance treatments for specific pediatric health issues and in the 1990s a new criteria, the imperative of pediatric research, was included to push for improved health outcomes in the pediatric population and to limit the off-label use of drugs in children[8]. Current guidelines specify that non-beneficial research can be conducted with children if risks are minimal or slightly above the minimum threshold, but have the potential to generate important results and do not expose children to procedures that they would not encounter in their daily care[14].

Changes in regulations from limited research to increased child participation have advantages and disadvantages. Progress in health outcomes and treatment of children was achieved, but guidelines may be too weak to offer protection in what concerns loose clinician interpretation of minimal risk and the development of an utilitarian approach in the study and diagnosis of some child disorders[13,15]. However, these drawbacks did not result in questioning the necessity of such non-beneficial research or calls for proposal of a better ethical justification in research with children[16].

Developing a non-consequentialist argumentation for research with no clinical benefit

Treating children only as means is morally wrong and efforts to show how children enrolled in non-beneficial research can be ends in themselves take into consideration either children's choices or their interests[17,18]. The first thesis is based on a will-approach that requires researchers to engage children's wishes. It presumes that present participation in research will match the decisions children would make later in life out of altruism. This view is intrinsically flawed as it is based on children's presumed or later capacity to consent and, despite empirical research to the contrary, suggests that persons will always act altruistically[16,19]. The second thesis is centered on children's wellbeing in relation to research risks[19]. Such analysis helps identify what should be an acceptable threshold risk in order for the research to be ethical, but does not address the children's contribution as social beings[19,20]. Several scholars who focus on altruism in children as a justification and means to benefit them are equally unconvincing. Evidence lacks on how such children may experience their participation and it cannot be assumed that young children with low cognitive capacities can even grasp the meaning of their contribution for others[18]. Therefore, the fostering wellbeing approach leaves unanswered the question why non-beneficial research should be allowed in the first place[16,19].

A contribution to a valuable project

Wendler proposes an ethical analysis based only on risks and benefits, emphasizing the acceptability of exposing some children to harm for the benefit of others within certain limits, as imposed by legal provisions[16]. This approach allows for the inclusion of the societal aspect of research and the identification and role of non-clinical benefits to research participants[19]. He contends that it is in the interest of persons to have a better overall life and that participation in research, viewed as ways to engage in community activities and build

meaningful relations, can help achieve this[19]. Furthermore, Wendler[16] affirms that children, even in the absence of capacity are contributing their bodies and that this benefit is only maximized for children with increased cognitive capacities[16].

The contribution to "a valuable project" fails in consideration of several aspects: not all children will cherish their participation in research as a beneficial contribution, and the research study may not reach the anticipated aim of producing important generalizable knowledge[21]. Furthermore, Wendler's focus on risk/benefit ratio[16] cannot identify why it is morally wrong when children from a particularly disadvantaged group are overrepresented in non-beneficial studies[22]. This issue is particularly important when analyzed in relation to Ramsey's absolute claim that all non-beneficial research with children is wrong[7]. In the end, children can achieve a meaningful life by making different contributions which do not necessarily involve exposing them to physical risks[18].

The body as subject

Both the will and the best interest approach described above take as a given the fact that children are vulnerable due to their incapacity to consent and their propensity to be harmed by research. However, all persons experience vulnerability in their bodies as it is an inherent condition of being human[23]. The likelihood of being harmed through the body is not specific to children or linked only to specific activities, such as pediatric research. As such, persons are not only vulnerable in their bodies, but are vulnerable to their bodies, as the person and her corporality form the whole human being[24].

Vulnerability in bioethics is usually interpreted as a weakness that needs to be corrected either through protective practices or by elimination[23,25]. It poses the danger to stigmatize some groups[26] and may not be able to recognize or be sensitive to different vulnerabilities that may concurrently act upon a person[27]. Correction as a remedy to vulnerability is usually

connected to emphasis on the power of informed consent and the role of autonomous decision-making, therefore acknowledging that persons can dispose as they see fit of their bodies[28]. However, people do not own their bodies, from which they cannot be separated and which are part of the person they are[29]. Furthermore, the vulnerability intrinsic to having a body cannot be eliminated. At the same time, persons interact with others through their bodies which facilitate building relations. As such, the body becomes an expression of the person and the means with which she engages with the world[28]. Particularly for children, this embodiment of the person is visible when, as their cognitive and emotional capabilities are developing, they use their bodies to communicate.

Understanding children's participation in non-beneficial research through the lenses of the body as expressive of the person shifts the focus from a passive contribution to an active one. As such, the body is not a mere object that, used to instrumentalize children, but a subject that actively contributes. This view resolves the problem of children's inability to provide consent and does not necessarily require a conscious knowledge of the symbiosis between self and body[30]. The relation body-self becomes evident in case of illness, when one experiences the harms of the corporeal being through pain and other physical symptoms[31]. What becomes evident in this situation is also the impossibility of eliminating the vulnerability given by the body, as the suffering cannot be transferred to another person.

Conclusions

A body-subject interpretation to children's participation in non-beneficial research contributes to finding a solution to the ethical dilemmas that this type of research poses in relation to minor patients and their vulnerability. Wendler's suggestion to acknowledge children as participants and not mere subjects of research hints to the validity of the contribution made through embodiment[19]. The embodiment-approach also incorporates the

additional non-clinical benefits that children may experience and supports the need for child's developmental assent and the importance of respecting dissent. However, it is not a standard to be applied practically, but rather offers a general defense of conducting non-beneficial research ethically.

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Participation in pediatric oncology: Views of child and adolescent patients

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Abstract

Being involved in healthcare can result in benefits for children and adolescents, but some patients may choose to have a reduced involvement in decision-making and information sharing. In order to explore children's perspectives on being involved in cancer care we conducted a qualitative study in Switzerland. Interviews with 17 pediatric patients were coded using thematic analysis. Participants' accounts described participation ranging from direct to indirect involvement, with more child input in general discussions regarding treatment and care. Patients were in the background of decision-making processes regarding treatment, but had their voice heard in matters regarding care delivery. Variation in patient involvement in care may account for children's and adolescents' preferences and fluctuating wishes while undergoing treatment for cancer.

Introduction

Children's participation in care can be achieved in different ways, most of which are part of communication processes: information exchange, expressing opinions, collaborating in making decisions[1,2]. The different degrees of involvement in care are influenced in pediatric oncology by prioritization of compliance with strict treatment protocols[3,4]. As such, children's preferred level of involvement may be difficult to achieve and result in increased loss of control, anxiety and uncertainty[2,5]. We conducted a qualitative interview study in Swiss pediatric oncology centers to investigate child and adolescent views on how they participate in care, their position and needs in communication.

Methods

Interviews with the three parties – parents, patients and physicians - involved in care for children diagnosed with cancer were conducted in Switzerland. For the purposes of this study we report data only on child and adolescent interviews. Collaborating physicians recruited families from 8 pediatric oncology centers from three linguistically diverse Swiss regions, Swiss-French, -Italian, –German. Purposive sampling was used for sampling procedures. After parental agreement, minor patients were invited to participate. A guide with semi-structured questions on experiences from diagnosis onwards was used to elicit children's opinions. Data was analyzed using qualitative thematic analysis.

Results

Interviews were conducted with 6 female and 11 male patients with ages between 9 and 17 years at the time of diagnosis. Discussions with participants and data analysis resulted in the identification of three themes of participation. The first theme on communication and decision-making roles revealed various positions that minor patients occupied at diagnosis

and then later in the course of treatment. These oscillated between being present in communications regarding treatment and hospitalization procedures and contributing to discussions or in making decisions. Direct involvement for most patients started with diagnosis disclosure. However, few patients did not witness the diagnosis discussions between parents and physicians. These children were later informed by their parents.

Children and adolescents described how in the course of treatment they started to participate in discussions by asking questions and through physician and parent invitation to contribute to making decisions. Accounts of involvement in decision-making highlight the fact that the patient's role was stronger in choices regarding care. In what concerned treatment decisions, children and adolescents were sometimes given the opportunities to express choices regarding research participation or fertility preservation. However, their position was not that of a final decision maker. Research participation decisions were made collaboratively with parents and physicians and fertility preservation choices were taken into consideration only when they did not delay treatment.

The second theme of participation highlights the effect that it had on children. In general, participants were satisfied with their participatory roles and with how communication was carried. However, some children identified difficulties in the way information was shared. These ranged from inability to understand, physician use of medical language, excess of information in relation to technical procedures (e.g., insurance schemes and payments) and insufficient explanations regarding medical tests and side-effects. This sometimes left patients feeling frustrated and anxious, particularly in the context of decision-making.

Participants' thoughts and opinions about participation is the third identified theme. Children and adolescents perceive involvement as the normal thing to do, as they are the ones having and suffering from the illness. Moreover, patients expressed a wish to receive information from the most competent persons, the physicians. Oncologists were perceived as qualified to

explain the illness and treatment and also patient-physicians direct communication can undercut parental strategies to limit information. Preferences for how communication should be carried varied among patients and even changed for some children at different times in the course of illness.

Discussion

Child and adolescent accounts of participation in cancer care emphasize the need to advance patient involvement in care. Similar to previous studies, participants in Switzerland described their involvement in communication and decision-making as convoluted[3,6,7]. They discussed difficulties with both involvement and non-involvement as shaped by parents' and physician's behavior. At the same time, children reported their own struggles to find a context dependent degree of involvement that is satisfactory to them. Their preferences to sometimes participate and other times to be less active in communication and decision-making emphasize the need to repeatedly assess their needs[5].

Hart's model of participation describes various involvement levels that are similar to those discussed by participants in the study[8]. The ladder has its range from a bottom child-initiated level to a model of collaborative decision-making placed at the top. The visual pyramid of this model suggests that at the higher levels the achieved participation of children is more important compared to the bottom levels[8]. However, involvement has a qualitative value and when in harmony with child abilities and wishes it is appropriate and meaningful. Different types of involvement can be better conceptualized by introducing a concept of degree of participation. This can account for various influences and burdens experienced by patients and should be individual based. As such, children may favor different degrees of participation and be satisfied with moving from one to the other depending on context and their specific

preferences at that time. The degrees of participation from less to more involvement are likely to meet children's fluctuating and developing needs[3,9].

Future studies should examine how clinicians can better identify and assess the needs and preferences of individual patients. Steps to facilitate communication among physicians, parents and children should be explored with the aim to improve patient comprehension.

Conclusions

Children's and adolescents' views are not commonly explored in clinical research. This study contributes minor patients' reflections on their care and emphasizes that participation is desired and meaningful for children with cancer, despite difficulties.

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"Dry your tears up, all your crying

Cannot fix me up my darling

(…) Twisty, turning winding path

I could listen to your laugh

As we tiptoe on these humble truths"

(Fix me up, Sammy Brown & Zach Sobiech[1])

Chapter IV

End-of-life care: controversies and ethics in search of a good death

[1]A Firm Handshake, Fix me up. Lyrics written by Sammy Brown and Zach Sobiech.

End-of-life decision making in pediatrics: literature review on children's and adolescents' participation

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Abstract

Children's and adolescents' abilities may support their involvement in decision-making at the end-of-life. The developmentally appropriate involvement can be ethically justified, but the outcomes of minor patients' contribution to make difficult choices when treatment fails are unclear. The purpose of this article is to offer a systematic review of pediatric studies carried at the end-of-life and investigate what are the effects of children's inclusion. Searches in five biomedical databases were carried to identify research with minor patients at the end-of-life. We screened overall studies for patient characteristics, choices that were made available and the outcome of final decisions. From 57 included articles, the majority examined family and clinician experiences and only 14 looked at child and adolescent views. Both positive and negative outcomes of participation were identified. These results emphasize a lack of research to identify factors that have a positive influence on child and adolescent participation at the end-of-life. Also, barriers and possible frictions between physicians, parents and patients are overlooked. This stalls progress on developing mechanisms to resolve eventual communication problems in decision-making processes and to support child participation at the end-of-life.

Introduction

Childhood cancer treatment improvements have reduced mortality rates[1]. However, for some patients therapy will fail, leaving families, children and physicians to make difficult decisions regarding enrollment in phase I clinical trials, opting for withholding or withdrawing of life support, trying aggressive chemotherapy or choose palliative care. Professional organizations in pediatrics propose a developmental approach to patient participation, which in practice should result in different levels of involvement, from being informed to being the main decision maker[2,3]. Minor patients' involvement is limited by legal provisions and should take into account parental and patient wishes for involvement.

Research on benefits of child inclusion in care at the end-of-life (EOL) suggests that open communication helps relieve anxiety and may results in improved outcomes for parental wellbeing. However, positive outcomes have not been weighed against negative effects of inclusion and there is significantly less knowledge on harms[4-6]. At the same time, patient preferences and needs of inclusion and communication vary in the course of illness and some children may resort to avoidance to protect themselves against distressing information[7-9].

The purpose of this review is to assess the involvement of children in end-of-life decision-making (EOL DM), the effects of their participation on all parties and on the process itself. It systematically analyzes EOL studies to identify ways in which children are involved and subsequent outcomes.

Methods

A three-phase systematic literature review based on the PRISMA[10] protocol was conducted to scan for original studies on child and adolescent involvement in EOL care in five databases: PubMed, Medline, PsycInfo, CINAHL, and Sociological Abstract. Research in neonatal care and on physician assistance in dying and euthanasia were not included.

Results

A majority of the 57 research articles selected at the end of the systematic review process were concerned with parents' or healthcare professionals' views and only 14 also captured the experiences of children and adolescent patients. Decisions offered to families and patients included: withdrawing/withholding of treatment, advance care directives, phase I clinical trial participation, continuous deep sedation, choosing place of death.

From the studies that had adult participants, some mentioned children's preferences, emotions or choices as being taken into account in decision-making[11-20]. A small number of articles described in more detailed how child participation was achieved and even fewer reported outcomes[15,20-23]. Parents stated that children sometimes were told about the adults' decisions to stop treatment[20] and had a gradual involvement in decision-making processes[23]. Participation was influenced by how much children understood what they were told, their individual awareness about what was happening around them[15,23] and cultural attitudes towards cancer[21]. The approaching of death was discussed rarely with children[22] and only in five studies some children were the ones to make decisions[17,24-27].

Studies that enrolled child and adolescents participants examined their views on being involved and their preferences for collaborating with adults or being the sole decision makers[28,29]. Six articles also tested the development of advance care tools or directives for minor patients and reported better agreement between child and parent preferences after implementation[30-35]. In one qualitative study patients were supported to explore their preferences and values regarding the illness and decisions around the time of death[29]. A report study on end-of-life decisions highlighted the burdensome effect and the distress these can cause in a patient as her condition deteriorated[36].

Discussion

Child and adolescent patients' views at the EOL are scarcely researched and the outcomes of their involvement in care are inconsistently reported and cannot be compared across studies. Parents' and clinicians' attitudes and experiences with child involvement are examined more often. Adults' accounts show that children's preferences influence decision-making processes, but it is unclear how and to what extent. Different aspects of participation were identified in the reviewed studies: offering information[37,38], engaging children and adolescents in planning future care[30,31,33,34], supporting patient expression of personal beliefs and wishes for when they die[23,36], seeking child agreement with what adults decided[20], communicating with patients about decision-making and collaborating with them[4,36,37]. However, some possibilities reported under child participation, such as patient consultation after the decision is made, may be viewed as barriers to involvement or parents' strategies to avoid discussing difficult treatment decisions with children. Despite these findings, there is research showing that teenagers wish to have a say and make decisions in collaboration with parents[28]. Several advance care planning tools prove to have clinical utility and facilitate EOLDM[30,31].

Conclusions

Minor patients' involvement in EOLDM is difficult, but gradual efforts to identify children's preferences can result in positive experiences by showing respect for patients and help them leave behind memories. Helping children and adolescents voice how they want to be remembered can improve medical and spiritual care and thus benefit patients and families. Future research should explore how to adequately and better achieve participation in EOLDM.

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Continuous deep sedation and euthanasia in pediatrics. Does one really exclude the other for terminally ill patients?

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Abstract

Debates on morally acceptable and lawful end-of-life practices in pediatrics were reignited by the recent amendment in Belgian law to allow euthanasia for minors of any age who meet the criteria for capacity. Euthanasia and its legalization in pediatrics are often opposed based on the availability of aggressive palliative sedation. For terminally ill patients this type of sedation is often identified as continuous and deep sedation until death (CDS). We demonstrate that this reasoning is based upon flawed assumptions: (1) CDS is a morally preferable alternative to euthanasia; (2) CDS can meet the same patient needs as euthanasia; (3) children lack the capacity and experience to make end-of-life decisions; (4) unlike euthanasia, CDS does not raise capacity issues. Our aim is not to reject CDS as a valid option at the end-of-life, nor to offer a clear-cut defense of euthanasia for minors, but to emphasize the ethical issues with both practices.

Introduction

Dying patients with refractory symptoms face physical and psychological suffering as symptoms are no longer controllable by standard therapies. To relieve their suffering, patients receive palliative sedation, that is, the monitored and intentional administration of sedative drugs to induce reduced or absent awareness (unconsciousness)[1,2]. Sedation can be mild, intermittent, superficial or continuous[3]. Professional guidelines consider palliative sedation an accepted and established form of care at the end-of-life (EOL) if patients are (1) terminally ill; (2) in the last stages of life and (3) suffering from one or more refractory symptoms[2-8] (see Table 1).

Continuous deep sedation (CDS) is the means to render the patient fully unconscious until the moment of death[i]. In addition to the above criteria, this sedation is accepted only for imminently dying patients for whom intermittent or respite sedation fails to provide relief[8,9]. Therefore, CDS has to be proportional, a last resort after other palliative means have failed. The proportionality principle guides medical procedures by balancing harms to the patient (suffering from refractory symptoms) with side-effects (reduced or loss of consciousness) and risks (potentially life-shortening). The greater the suffering, the higher the risks physicians can take[10]. It is important to emphasize that CDS is a last resort measure which requires the consent of the patient (or surrogate)[9]. When these two additional conditions are met, most institutions do not require a review process by another physician or to report the procedure to institutional or health authorities[2-4,6,7]. Some organizations suggest an interdisciplinary consultation prior to starting CDS[4,6,7]. However, there is no agreement on the type of experts that should be involved and whether the consultation should result in consensus (see Table 1).

TABLE 1. International guidelines' different provisions on the use of continuous deep sedation (CDS) in terminally ill patients

Associations' and institutional Guidelines	Whom it addresses	Survival expectations at the time of administration	Requirement for Consent	Safeguards for CDS use*
National Hospice and Palliative Care Organization (NHPCO), 2010, US	Patients – no mention of age	14 days	__ (implied: "Only (the) patient can identify when suffering has become intolerable")	ANH[a] decisions are independent of the CDS decision Judicious use of medication following evidence-based clinical protocol and ongoing monitoring Use by clinicians experienced with medications and type of sedation Interdisciplinary case by case consultation with experts in pharmacology, pain and symptom management, interventions for suffering (psychological, interpersonal, spiritual, and other deemed relevant by individual patients) Patient and family-centered care Clinicians' ongoing education in symptom assessment and management, ethical considerations related to use and family-centered care Institutional regular and formal review of utilization, policies and practices
European Association for Palliative Care (EACP), 2009, Europe	Patients – no mention of age	hours or days, at most	YES	Early on individual discussions, contingency planning and periodical patient and family goals assessment Formal documentation of discussions ANH[a] decisions are independent of the CDS decision Evaluation by clinician with experience and expertise in palliative care; if possible, interdisciplinary evaluation Use of validated prognostic instruments If possible, physician and nurse start administration together Clear patient assessment procedures; the only critical parameters are comfort ones Midazolam recommended as usually used

Royal Dutch Medical Association (KNMG), 2009, The Netherlands	Patients – no mention of age	1 to 2 weeks	YES (if patient lacks capacity, the patient's representative consents; patients partially lacking capacity should be involved as far as possible)	Establish that symptoms are refractory beyond reasonable doubt and using good medical practice ANH[a] decisions are independent of the CDS decision Have a criteria for initiation, consultations with patient and/or family and specialist Keep record of all decisions and interventions Physician presence at initiation Recommended drug: Midazolam; the use of morphine is considered bad practice Mandatory consult (preferably palliative care specialist) when clinician expertise is insufficient, patient life expectancy is above 14 days or uncertain
University Hospitals of Geneva (HUG), 2009, Switzerland	Child and Adolescent patients	days and no more than few weeks	YES (patient oral assent and parental oral permission) depending on child's age and capacity	Follows EACP[c]'s recommendations and extends them to minor patients Expert case by case interdisciplinary discussions that result in consensus Early on discussions regarding treatment, goals and palliative sedation options Children's participation in discussions and decision making should be supported, according to age and capacity for 2 reasons: the determination of intolerable symptoms requires the patient's participation sedation effects are significant: inability to communicate with the family Protocol including the frequency of monitoring, observation of comfort Documentation of indications, discussions with patient and family and their agreement, goal, all decisions and interventions, patient, family and provider satisfaction

Report of the Council on Ethical and Judicial Affairs, AMA[b], 2008, US	Patients – no mention of age	in the final stages of terminal illness	YES and/or patient's surrogate informed consent	Document the rationale for administration Multidisciplinary team consultation, including an expert in palliative care Implement monitoring process of appropriate care upon administration

* Commonly, all Guidelines include safeguards referring to patient's pre-conditions: (1) terminally ill; (2) imminently dying and (3) suffering from one or more refractory symptoms. Additionally, a step-by-step approach in which the level of sedation should be guided by the level of consciousness reduction required to sufficiently relieve symptoms is generally recommended.

[a] ANH – Artificial nutrition and hydration

[b] AMA – American Medical Association

CDS administration is recommended after careful consideration of the adverse outcome of taking away consciousness, and the risk of potentially shortening life[2]. These negative effects are condoned because CDS is considered to be the only means to alleviate patient's suffering. Clinicians seem thus to be operating under the doctrine of double effect (DDE), which plays a central role in justifying and permitting certain practices at the end-of-life when the physician (non-)intervention brings about both good and bad effects[11]. The DDE's classical interpretation originates from the Roman Catholic moral tradition. It has often been used to solve moral dilemmas regarding end-of-life practices[11] and formulates that an agent's (physician's) actions are licit when 4 conditions are met cumulatively, at the same time: (a) the action, in itself, should be positive (good) or at least neutral; (b) the action intends to achieve the good and not the bad effect; (c) the good effect cannot be achieved through the bad effect and (d) effects must be weighed so that only a "proportionally grave reason"[12] allows for the bad effect to happen[13]. In CDS clinicians' aim is not to hasten death, but to provide relief in the face of the anticipated, parallel, though *unsought*, consequence of possibly shortening life[14-16]. Therefore, the good, intended effect is to alleviate pain and the bad effect is the loss of consciousness, also sought, but licit only when the proportionality principle is respected and there is a valid informed consent, as explained above.

Hastening death in CDS and survival time in patients receiving palliative sedation is a highly debated issue[9,17-20]. Studies correlating time of death and palliative sedation[21-23] report that there is no evidence that sedation significantly hastens death: "sedation has no apparent effects on patient survival, at least on a mass level"[23]. Only for a limited number of patients, palliative sedation is associated with catastrophic events, causing death[23]. In light of such conclusions it seems that DDE's use to justify CDS is superfluous.

A systematic literature review[24] concludes that evidence provided by studies should be interpreted with caution given the high bias risk due to lack of randomization. Most studies

fail to provide adequate description of the timing, duration and level of sedation[24]. In these clinical trials a comparison is made between non-sedated and sedated groups, but the degree of sedation is generally not further specified (mild, intermediate or deep and continuous or intermittent). Thus, the impact (or lack thereof) of CDS, on the time of death remains difficult to ascertain. This difficulty also stresses the need to use terminology accurately. Palliative sedation includes a spectrum of different interventions that might evolve from relatively light sedation to CDS toward the end of a patient's life. While it is important to recognize the proportionality principle (correspondence between symptoms and level and length of sedation) and not to equalize CDS with palliative sedation as such, it is equally important not to "hide" or "obscure" CDS under the umbrella term of palliative sedation. Even if CDS "rarely if ever hastens patient death"[10], it takes away an important human good, consciousness[9]; a dying patient might claim that his "real" life is ending when he loses consciousness following CDS. It is this particular context that needs to be considered when discussing the DDE.

In ethical discussions around CDS the DDE plays an important role, but it is also criticized. Not only it is impossible to know what is in a physician's heart and mind regarding hastening death as intentions are often ambiguous and contradictory[25], but the fact that loss of consciousness is the means to the good effect (pain relief) and thus not just an *unintended* negative side-effect, significantly undercuts the force of the entire DDE argument as it is clearly in opposition to the third condition (c)[9,12].

Despite difficulties in applying DDE to CDS, guidelines rely on it to draw a clear distinction between the palliative scope of CDS and the more direct life-shortening intention of other EOL practices, such as physician-assisted suicide (PAS) and euthanasia[2]. PAS refers to the practice of prescribing drugs in lethal dosages by a physician with the purpose of providing patients with a means to end their life[26]. Voluntary active euthanasia (VAE)[ii] is the practice

of a physician intentionally ending a patient's life upon the voluntary and persistent request of that person. In those (few) countries where PAS and VAE are lawful, a review and report to health authorities are mandatory[27].

Recently, the debate on morally acceptable and lawful practices at the end-of-life has been reignited by a change in the Belgian law to allow euthanasia for minors of any age, as long as they meet the criterion for capacity[28]. Critics of this bill argue that euthanasia is morally wrong as the physician's aim should always be to relieve suffering, not to end life. In their view, euthanasia is not only incompatible with physician's role as healer, but also the availability of aggressive palliative sedation or CDS eliminates the need for the legalization of euthanasia as it can alleviate even extreme suffering[29]. The main purpose of this article is not to reject CDS as a valid option, nor to offer a clear-cut defense of euthanasia, but to overcome the deadlock in this debate by raising awareness about the ethical issues with *both* practices. Decisions at the end-of-life are loaded with uncertainty and require demanding clinician responsibilities in guiding honest communication for patients approaching death[30,31]. Death and dying, and EOL options often incite many passionate reactions in the public sphere that overshadow the personal experiences of dying patients. Despite advances and significant financial and structural efforts, palliative care services are still underdeveloped and care does not always reflect patients' wishes on how to die[32]. Resisting euthanasia by proclaiming aggressive sedation to be a "panacea" ignores the shortcomings it may have in eliminating various forms of physical and existential suffering[33]. That leaves little room for discussing what patients envision as a death without suffering, commonly referred to as a *death with dignity*. Also for minor patients dying under sedation may infringe upon their dignity when this process does not correspond to their wishes for a "good death". It may circumvent patient needs and be disrespectful toward children who have the capacity and experience to express such wishes. The dilemma for these patients and their families is not if they will die, but *how*

they will die. We believe euthanasia and its legalization in pediatrics should not be opposed based on the availability of CDS because this type of reasoning is grounded upon 4 flawed assumptions: (1) CDS is a morally preferable alternative to euthanasia; (2) CDS can meet the same patient needs as euthanasia; (3) children lack the capacity and experience to make end-of-life decisions; (4) CDS does not raise capacity issues.

1. Is CDS a morally preferable option?

The practice of palliative sedation ("to the point of causing unconsciousness and hastening death") has been used in courts in North America as an argument against the need for legalizing PAS. In court decisions the two practices are placed side by side, separated by a lawfulness criterion and the seemingly arbitrary proclamation that the former is morally preferable to the latter, as long as it operates under the DDE[34]. This means that even if the life-shortening effect of CDS might be rare, its potential to hasten death is not fully excluded as the strict rules under which sedation to unconsciousness is condoned derive exactly from accepting this possibility. This also means that the intention of the moral agent, i.e. physician, cannot be overlooked in the ethical evaluation of CDS.

The "preferable alternative" (PA) argument has been criticized[18,20,35] by focusing among others on physician intent, informed consent and the so-called natural death hypothesis[20,36]. The same PA argument has been used in opposing euthanasia legalization in children in Belgium[29]. Therefore, we briefly recast this ethical discussion by focusing on CDS and euthanasia within a pediatric context.

Studies investigating CDS, mostly come from European countries (Belgium, the Netherlands, United Kingdom)[37-39], but also Asia[23]. This research shows that an intention to hasten death might be either concomitant or explicit to physicians' reasons for starting CDS[37-39]. The one study conducted in pediatrics suggests that this might also hold true for

CDS in children. For a quarter of minor patients in a study in Flanders (Belgium) hastening death was concurrent to or the aim of administration[40], eroding thus the apparent distinction between CDS and euthanasia. Contrary to the stipulated guidelines, almost none of the children had requested or given consent for CDS and in over 20% of the cases parents' consent for deep sedation was missing[40]. Research also seems to indicate that the use of CDS has increased in end-of-life care[21,37,38,41,42]. This seems to be in contradiction with the "last-resort" framework within which CDS should operate. This reported increased use may only concern a small series of studies that are in no way generalizable to the use of routine palliative sedation or of CDS in all contexts. Also, the ethical evaluation of a practice such as CDS cannot be based solely on its possible "abuse". However, these studies highlight that both patients' and physicians' true intentions are difficult to assess. Goals and motives are not only multiple[25], they are also often the expression of unconscious volition. Hence the DDE does not constitute a reliable moral basis to distinguish CDS from other EOL practices[17]. In practice, the cumulative conditions for DDE are difficult to apply and adherence to moral norms may be inconsistent when considering the two bad effects, loss of consciousness until death and the potential to shorten life.

The increased frequency use might also indicate that CDS is becoming more and more acceptable among patients, family members and physicians. According to Raus and colleagues[36] increased acceptability has something to do with the growing association between CDS and the concept of a "natural death", where nature determines the timing of death as opposed to a so-called medicalized death. To patients and their families, CDS may offer comfort as it resembles a deep sleep in which patients gradually fade away due to their illness and without apparent direct human interference. In the case of children these elements are even more likely to be sought, as it is particularly difficult for parents and care providers to face such losses[30,31,43,44]. However, CDS is only a mimicry of a natural death as the patient

is not sleeping, but in a medically *induced* coma. Death is not automatically the result of the underlying disease, but can be caused by the unpredictable effects of sedation, such as respiratory depression and aspiration[2,45]. Besides that there is not a clear correspondence between CDS and natural death, this association is also deeply problematic as it minimizes the ethical problems of CDS and sidesteps the debate about its ethical safeguards. Natural, in fact, is often equated with being morally good and therefore upheld as a normative ideal. Depicting CDS as "natural", signifies portraying CDS as a normal and acceptable practice as opposed to deaths caused by other end-of-life practices, such as euthanasia[36]. However, as might clear from the above, in practice this ethical difference is not as clear-cut as often presumed because it is not always easy to determine whether a physician aims exclusively at the relief of suffering or also at the hastening of death. Moreover, if CDS is practiced without patient consent, it has an element of non-voluntariness and possibly becomes more problematic than euthanasia. Hence, it is highly problematic to oppose euthanasia by arguing that CDS is an ethically preferable alternative because in practice the standard ethical norms for CDS are difficult to apply and often violated.

2. Does CDS meet the needs of all dying children?

Patients with terminal illnesses experience many physical and psychological debilitating symptoms and may express different wishes at the end-of-life. For some patients, living with a poor prognosis and declining health prompts them to make a request to die with dignity[27,46]. Research shows that those who are granted euthanasia are often younger (below age 65) and highly educated patients. They experience both physical and existential (fear of loss of autonomy and quality-of-life) suffering, in the whole of their person[47]. CDS is more frequently used in elderly patients (over age 65)[27,48] who are near to death and have unbearable physical pain combined with other refractory symptoms, such as dyspnea and

anxiety[37-39,42,49,50]. CDS use is generally not accepted for the only purpose of relieving existential suffering[10].

Legal regulations portray a similar image: in countries or states where euthanasia is legal, it is an option for patients who seek a dignified death on their own terms in order to avoid unbearable physical or psychological suffering[51-53]. In the Netherlands and Belgium, euthanasia is allowed independent of a narrowly defined expected survival time. CDS on the other hand, is indicated for imminently dying patients with intolerable physical symptoms that have proven refractory to standard treatment[2-4,9]. Hence, CDS and euthanasia seem to meet distinct needs of different kind of patients. Therefore, it would be wrong to preclude considering euthanasia a priori by referring to the CDS option.

In pediatrics, the criterion for CDS administration is more rigidly applied considering the limited evidence base of palliative sedation in this population. Due to the relative lack of data, administered doses could be too high and increase the risk of hastening death to a greater degree than necessary[40,54,55]. As a consequence, CDS in children is usually used very restrictively and started only in the week or even a few days before death. Although withholding or withdrawing medically provided fluids and nutrition is consistent with symptom relief at the end-of-life, for pediatric patients they are generally continued[40]. This shows the difficulties pediatricians face in limiting therapeutic treatment when confronted with children at the end-of-life. It may also explain the general concern and disquiet about the Belgian Parliament's decision to allow euthanasia for chronically ill children.

In Belgium euthanasia has been legal since 2002 for persons 18 years or older, but since March 2014 the age limit has been removed. Euthanasia is considered an option in pediatrics if patients are terminal and experiencing constant and unbearable suffering. Other criteria include: an explicit and voluntary request made by the minors themselves, consent from both parents and a multidisciplinary team should examine the children's decision making capacity

for discernment. Therefore, children with an intellectual disability or mental illness are excluded from opting for euthanasia[28,56]. Moreover, unlike for adults, the Belgian law excludes *existential* suffering as a *self-standing* criterion to request euthanasia in children.

Despite these criteria, the recent amendment has been fiercely opposed not only because of its possible misuse in a vulnerable population such as children (the so-called "slippery slope" argument), but also because it is presumed that the request for euthanasia by children cannot be motivated by the same "sophisticated" needs that adults express in their requests[29]. Children can be vulnerable to unbearable pain, but they do not have enough life experience to understand death or to be susceptible to fear of humiliation or loss of control that end-of-life conditions may bring about. Their physical suffering can be relieved by aggressive palliative sedation.

For patients sedated to unconsciousness until death, it is impossible to know if their pain and suffering was alleviated, as no means of self-report are possible. However, observational studies on the effects of palliative sedation at the end-of-life produce limited evidence of palliative sedation's efficacy in adequately controlling symptoms as patients showed signs of fear and anxiety[23,24,42,57]. This should be considered worrisome, especially in pediatrics where pain management is a sophisticated issue.

Research shows that, compared to their healthy peers, children with terminal illnesses often have a mature concept of death and develop an understanding of how they want to die and how they want to be remembered[58-60]. Some minors even express the belief that "life on a machine is not living"[61]. Some children may want control over what is happening and to enjoy life as long as possible [58,61]. These considerations about death and dying mainly come from their illness experience, the familiarity with the medical context and their interactions with health care professionals and parents[62-64]. Hence, we should ask ourselves whether CDS can really meet the needs of all dying children or whether such an assertion is the expression

of an adult-centric view that circumvents knowledge gained on children's understanding of illness and death as described by several scholars[59,60,65-67].

3. Children's capacity for end-of-life decisions

Closely connected to the needs-discourse is the one concerning capacity. Opponents of euthanasia in pediatrics, not only argue that children lack so-called "sophisticated" needs, they also deny that children have the capacity to make legitimate requests for euthanasia[29]. This view seems to be grounded in the widespread belief that children are impaired and inexperienced and adults know best. The problem is that such generalizations keep us from acknowledging what children can really achieve. Moreover, it sidelines the ever growing and significant body of international pediatric guidelines that seek to recognize children as moral agents capable of making decisions regarding their own health, at a level commensurate with their cognitive and emotional abilities[4,68,69]. Observance of this recommendation requires a careful assessment of the individual child's capacity in order to achieve involvement that is adequate and desired by the patient[70]. However, it would be wrong to claim that children's inclusion amounts to them dominating the decision process. The pediatric setting is unique as it is characterized by triadic discussions, involving health care personnel, children and parents. As such, physicians and parents should not only pursue children's best interest, but also facilitate decision-making by equipping children with the abilities to form and express their choices and wishes. Decision-making is influenced by personal values[71], which weigh heavier when making end-of-life decisions. One of the arguments put forth to object awarding decision-making capacity even to those underage patients who meet the criteria for understanding and reasoning is related to their developing values. In order to provide assent or consent to treatment, patients have to be able to appreciate and apply the information in relation to their own context and values[72,73]. Children often do not hold a stable set of values

until they reach adulthood or preferences and wishes fluctuate with age and depending on the decision to be made[74]. However, in the case of children facing terminal illness the change or maturation of values is cut short and the present held beliefs should be properly acknowledged[75]. This is especially the case in end-of-life situations when curative goals are no longer an option[76].

The transformation in goals of care and re-orientation towards quality of life and a painless death should incite clinicians and parents to initiate discussions on end-of-life issues and ask questions regarding minor patients' views, wishes and preferences. The use of advance directives can successfully foster communication between the three parties[66,77] and reveal children's wishes for funeral arrangements, the image they leave for others to remember, dying surrounded by family and last opportunities to say goodbye[62,78,79]. The Belgian euthanasia law is very much in line with these pediatric guidelines and triadic context as it requires not only the explicit and repeated request of the child, but also the consent of both parents and physician assessment of capacity discernment. Still, since there are no standardized methods to assess children's capacity[80,81], health care professionals mostly have to rely on their intuitive judgments on whether and how to apply these guidelines. The uncertainty about children's capacity may explain the protective attitude many people continue to have towards children and why the Belgian law may be disturbing to them. However, this difficulty does not clarify why CDS would be a morally "safer" option in pediatrics than euthanasia. This brings us to the fourth and last assumption of the preferable alternative argument.

4. Doesn't CDS raise capacity issues too?

The pediatric guidelines regarding child participation also regard palliative sedation. This means that also in the case of CDS, physicians and parents should involve children and seek

their consent. However, if we assume (for the sake of argument) that children are incapable to consent to euthanasia, then how could they possibly consent to CDS? What do opponents of euthanasia intend when they assert that children's request for euthanasia is not legitimate and CDS is ethically "safer"[29]? They seem to suggest that CDS does not raise the same issues regarding capacity as euthanasia because sedation does not involve any "sophisticated" choice[82]. Children merely consent to the ending of unbearable suffering and not to the hastening of death. Still, it is only thanks to its growing association with the concept of a natural death that CDS *seems* to involve less complicated choices. We should not forget, however, that CDS is a medically induced coma which leads to a complete loss of consciousness and amounts thus to a kind of social death[35]. This is why we should not merely condone the fact that in many cases the decision to start CDS is taken either by the parents and the physician together or only by the physician without asking the child's consent. Since the intent is not to kill, but to relieve pain, CDS might seem compatible with parents' and physicians' roles as respectively protectors and healers, but, although CDS does not kill the patient (or at least not in principle), it takes away consciousness. It suppresses sentience up to the point of passing away and thus entails a complex choice at the EOL. Some children may wish to express the desire not to spend their final days fully sedated. As such, CDS raises the same issues regarding decision-making and capacity as euthanasia.

Conclusion

In the literature euthanasia and its legalization in pediatrics are often opposed based on the availability of aggressive palliative sedation. We have shown that this argumentation is unsound as it is based upon 4 flawed assumptions. First, the ethical difference between sedation and euthanasia is not as clear-cut as often presumed given the uncertainty about physician's intent and the frequent absence of pediatric consent[9]. Secondly, as a measure of

pure pain relief CDS cannot meet the needs of all patients. Euthanasia choices alleviate not only physical, but also existential suffering. Relief from existential suffering may be especially valued by children who suffer from loss of control as a result of the experience with the illness and long hospitalization. Third, although children's capacity remains difficult to assess, pediatric patients, especially those who are severely or long-term ill often possess the abilities to form and express their choices and wishes. These capacities develop due to their ongoing interactions with physicians and parents regarding their illness and care, including at the end-of-life. Fourth, like euthanasia, CDS raises capacity issues since it is not just a way to alleviate suffering, but a medically induced coma to which children should give their consent. Still, as argued in the introduction, our aim was not to reject CDS as a valid option at the end-of-life, but to express our concern about the way CDS is used to cut off the debate on euthanasia in pediatrics as it not only precludes an open discussion on what might be a legitimate end-of-life choice for a certain type of patients, but it also keeps us from acknowledging the persisting ethical issues regarding the practice of CDS itself.

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[i] This paper looks into the ethical issues associated only with the practice of CDS that renders the patient unconscious and keeps her in that state till the moment of death. The act of continuous deep sedation is also referred to as terminal sedation, sedation to unconsciousness, end-of-life or total sedation. These terms may be used interchangeably in the text, but only to refer to the practice of CDS as described above.

[ii] Throughout the text we use the term euthanasia only to refer to the practice of voluntary active euthanasia, as opposed to withdrawal or withholding of treatment (passive euthanasia) or actions that end a person's life without the request and consent of that person (non-voluntary euthanasia).

Discussion

Discussion

Families' and patients' experiences of a cancer diagnosis are unique and there is little to prepare them for what comes after the initial disclosure[1-4]. Parents' needs and wishes for information and on how to have it delivered vary and indeed can fluctuate within one person during cancer treatment[1,3,5-8]. Some require more support in dealing with their emotions and, on the contrary, others feel disinclined to discuss or express feelings and fears, and prefer to focus on medical facts[8-10]. Physicians experience the toll of being the messenger of bad news and face challenges to identify what is the adequate information to share with individual families and patients[11-18]. Children's own needs and preferences and, above all, the suitable, effective clinician's and parent's actions to uncover their wishes make communication even more complex[13,19-22]. This work delves into the experiences of 35 parents and their children's oncologists, their descriptions and takes on communication and decisions about treatment and care. By analyzing participants' accounts against professional guidelines[23-26] and ethical principles[23,27], the doctoral work identifies different contexts - disclosure of diagnosis, treatment and care decisions, poor prognosis and relapse - and the ethical issues surrounding communication in these particular situations. Furthermore, the thesis then uses the knowledge gained from the empirical project to theoretically explore questions about communication in two other instances - end-of-life care and participation in research with little prospect of clinical benefit. The analysis is carried out by reflecting on ethical concepts of disclosure, parental rights and obligations, shared decision-making, physician responsibilities[12,15-17,28-30] and, more broader, their application in pediatric oncology taking into account diverse individual preferences. Recommendations on how to curtail communication breakdowns and how to improve the provision and sharing of information between parents and physicians in order to positively affect discussions with children are drawn. As the difficulties identified in

communication for children suffering from cancer may be common to pediatric chronic and life-threatening illnesses, the final guidance is relevant to various pediatric specialties.

Communication in pediatric oncology

Parents of children diagnosed with cancer are susceptible to vulnerability when they enter the medical setting, which for most is an unfamiliar environment. Patients and even parents, who may interiorize their children's symptoms[31], are rendered vulnerable by the condition of being ill and receiving arduous treatment. They, though perhaps to different degrees, experience a loss of control and dependency on the medical team[31-41]. For patients, vulnerability stems from young age and diminished maturity and dependence on others[27,31,42-44]. One of the main sources of vulnerability for parents comes from the reliance on others to provide them with information in order to care for their children[2,30,32,37,45]. Physicians and other medical staff are charged with providing adequate information in a helpful manner[24,25,39,40,43]. Many times clinicians are in the position of being messengers of bad news and as such the information they provide startles parents regarding their future[15]. Medical communication is essential for delivering optimal care, tailoring discussions to individual needs and for meeting ethical standards of informed consent[24,26]. Physicians' duties of informing parents and patients are complex. Therefore, communication in pediatric oncology cannot be subjected to a simple analysis and it requires a comprehensive understanding of the factors influencing all physician-parent exchanges, starting with the *day one talk*[2,11,40].

Diagnosis disclosure - the ethics of "truth telling"

Disclosure of diagnosis is at the cornerstone of setting the stage for building relationships between patients, physicians and parents and ensuing communications[5]. It is likely it takes

place when the family meets the treating oncologist for the first time and on the background of parents' previous experiences with healthcare personnel[5,46].

A cancer diagnosis disclosure model

According to professional guidelines communicating the diagnosis of cancer to families and patients must be based on complete information provision[23,25]. In practice, initial disclosure of diagnosis also identified in Swiss and Romanian pediatric oncology units is usually a private discussion between parents and physicians[40-42]. It is often structured around medical facts regarding the disease, additional needed tests, treatment and chances of restoring health, as well as common side-effects[5,40-42]. Additionally, parents' questions regarding how and when to tell children are being addressed[13,20,21,40]. However, there is no specific framework on how to deal with the myriad of parental predilections for delayed, partial or for non-disclosure to children. Guidelines mention flexibility to meet families' and patients' preferences and to accommodate different perspectives on disclosure[23,25]. This apparent contradiction between an open model of disclosure and suggestions to honor wishes to restrict information provision creates ethical and possibly legal dilemmas for physicians[14,40,44]. Many parents decide to be open with their children from day one, others may be encouraged by the first discussion with the physician that disclosure of diagnosis is necessary and appropriate[40,41]. Nevertheless, some parents are reluctant to reveal the exact diagnosis to children and some categorically refuse to[20,40,47]. These scenarios, although not the norm, are the ones most likely to uncover the ethical issues in cancer care communication and the difficulty to identify what physicians' responsibilities are and toward whom.

Professional duties to disclose

Physicians have a double duty when providing information about cancer, medical and ethical. This duty has to be fulfilled in relation to several actors: parents, the patient and other family

members, sometimes including children's friends and teachers[5,14,48-50]. From a medical point of view, deontology requires oncologists to give families and patients information regarding the diagnosis, the nature of the illness and its treatment[24,25,27]. Physicians have to use appropriate language that would ensure adequate understanding of medical facts and sufficient knowledge for decision-making[24,25].

From an ethical perspective, information sharing has to be individual based in order to support requirements for informed consent while be respectful of personal wishes and circumstances. Clinicians' ethical responsibilities extend also to reducing risk of emotional harm that can impinge on comprehensiveness essential for informed consent[24,25,51].

Responsibilities toward parents and patients

When they receive the news that their child has cancer, parents experience a carousel of emotions dominated by fears, need of protection and perhaps even denial[5,20,32,40,52]. Parents cannot claim a right not to know the diagnosis, its risks and treatment information as they are, under most jurisdictions, the ones designated to make decisions for their children[24,25,44]. As such, any distress that results from communicating essential medical facts is unavoidable and will inescapably affect negatively patients' and parents' notions of their lives in the present and future[53]. However, physicians have a duty to tend to individual parents' needs and show empathy and compassion in delivering such news[8,11,54]. Moreover, they have to assess what parents want to know and whether it is advisable to discuss cancer related issues in detail during the day one talk[2,25]. Information sharing may be titrated, broken-down to steps to allow respite from difficult news in consideration of parents' psycho-social state and if deemed in their interest. Such steps of controlling information provision are justifiable when they do not become barriers to delivering life-saving treatment or put at significant risk the health and wellbeing of the patient and parents. Similarly, physicians need to approach

parents regarding how and when to inform children[14,25,40], explore patient wishes and respect a child's expressed right not to know[25,55].

The benefits and harms of communicating openly, from disclosing the cancer and its risk, to prognosis and including the possibility of dying from the disease, are not clear cut[19,20,32,36]. Nevertheless, any anxiety resulting from knowing must be weighted with harmful effects of not being told anything or not much[13,20,32]. Moreover, patients hold different views regarding what is sufficient information and who should communicate it and how[21,22,41,42]. Given the multitude of factors and the uncertainty regarding benefits of full diagnosis disclosure to particular patients and their wishes, physicians should not escape reflecting on and giving parents' requests for limiting information significant weight[40,56].

Communication and decision-making

Parents appreciate when clinicians take time to discuss their situation and show caring behaviors, even if only by spending a bit more time at the bedside. Showing availability and doing more than just prescribe treatment, order tests and talk about medical results is comforting for parents[21,39,41,43,56]. Similarly, patients, particularly adolescents wish and need to establish personal relationships with medical professionals, as their own social network becomes limited during the treatment[18,42,43].

Parents in many centers share responsibility in caring for children and play a crucial role in ensuring compliance[32,39,41,56]. They appreciate being recognized as experts[5] and eliciting their views regarding care and treatment regularly it can render clinical benefits - improved understanding of medical facts and adherence[43]. Parents and patients alike get acculturated to the medical setting and if supported can take charge of many aspects of cancer care, such as prompt side-effect detection, keeping track of symptoms and what changes improve or aggravate them[39,41,56].

Understanding of information is one of the main elements necessary for providing parental permission for treatment[24,26]. Leaving parents with factual medical information that is difficult to sort out or asking them to make decisions for which they judge themselves as not qualified to make poses barriers to the decision-making process[39,43]. Adequate information provision is a mandatory step in obtaining informed consent for treatment and ensures that parents are equipped with the abilities necessary for making decisions. Just asking parents to make a decision, without assessing how they received the provided information is not fulfilling ethical standards or requirements for informed consent. Parents sometimes report that they cannot make a choice despite being told and explained what the treatment options are or when they would prefer more physician input. They need more time to process all relevant medical and personal facts before they can judge what is best for their child and their family[39,56]. Cancer treatment often is a context in which parents assess the situation as a "choiceless choice", having a predetermined decision[39,43,57]. At the same time, decisions during cancer care do not bear the same level of difficulty. Parties need to be able to negotiate their roles and, as the Swiss-Romanian studies show, sometimes even wish physicians to partake in the authorization of treatment[40]. This fact is surprising and goes against mainstream interpretation of autonomy and its role in decision-making. Unless encouraged by parental wishes, oncologists can sometimes allow too much autonomy for parental decision-making which may not be desired or can even be seen as a burden by parents[40,41,58].

Communication for research participation

Treatment in pediatric oncology is uniquely interwoven with innovative approaches and this adds another layer of complexity to parent-physician communication[59-61]. Permission for research participation requires additional detailed discussion of research protocols and therefore supplementary informed consent conferences and documents[24,39,62]. For parents the

conflation of standard and innovative treatment leads to difficulties in understanding multiple therapeutic options and randomized controlled trial (RCT) procedures[62].

Physician communication and limitations on understanding

Parental comprehension and uptake of information are related to several physician actions and attitudes[39,41,59]. Clinicians' ability to build rapport and partnerships, lack of skills and behaviors that fail to engage parents' active participation, as well as the content of the discussions physicians carry about children's enrollment in RCTs are associated with misunderstanding of fundamental research aspects[59,62,63]. Concepts such as randomization and voluntariness are complex and difficult to understand without some medical knowledge[39,41]. Physician use of metaphors, not regularly offering visual aids or failure to provide written research information, as well as clinician's omission to invite questions during informed consent conferences are contributing to low comprehension[59,62,63]. Research shows that employing figures of speech to describe treatment being allocated randomly does not improve understanding. Even more worrisome, many of these parents are more likely to enroll their children in studies compared to parents who have a good undertanding[62]. Understanding is poor also for voluntariness and parents often report feeling compelled to agree to participation[62].

Parents' ability to understand is also potentially weakened by the additional issues to consider in the care of their children[39,41,43]. The informed consent conference regarding research can be an additional stressful factor that impinges on ability to retain and comprehend information in a short period of time[62,63]. Understanding is also associated with socio-economic factors and is lower for parents belonging to minority or ethnic groups[62].

Ethical issues in research with chances of benefit

Difficulties in communication about research can results also from the fact that clinicians' role can become blurred when combining physician and researcher duties[60]. Countries have varied policies regarding research protocols aimed at testing improvements in treating pediatric malignancies[39]. While some national oncology groups regularly adopt new or innovative protocols to be used for all patients without proposing alternatives or the standard treatment, other offer treatment on various research protocols[64]. These approaches beg the question of what is research and how to adequately inform parents and obtain informed consent. When research is conducted with the prospect of benefiting patients and physicians are under clinical equipoise, it can be appropriate to propose parents an innovative treatment as a sole choice. For some parents this step can be legitimate, as it leaves them with less complicated decisions to make and follow physician expertise[39,41,43]. However, as Kodish cautions, meaningful informed consent needs to emphasize the risks, benefits and treatment options[64]. Parental capacity to comprehend is based on information provision and is essential for safeguarding the interests of the child and obtaining a valid informed consent[62,65].

Communication about non-beneficial research and other end-of-life options

Parents of children suffering from cancer are sometimes proposed to allow enrollment in phase I protocols[66-68]. These offer little or no direct benefit to individual patients and raise additional ethical challenges. Patients invited to participate have exhausted curative treatment options and decisions at this stage can also include limiting treatment or end-of-life care[69,70]. This begs the question of the suitability of inviting such patients to participate in research with little or no benefit[71].

Parental understanding of phase I clinical trials and physician communication about procedures and study aims are limited[61]. Phase I studies are designed to study safety in

children and not new treatment efficacy. Physician duties toward parents and patients should focus on protection and highlight the necessity of developing an ethical justification for such research[71]. Informed consent is a first requirement for morally allowing non-beneficial research with children[72]. Therefore, physicians' efforts should focus on improving communication with parents[61,73,74]. This can be achieved through education about research, mentioning of phase I clinical trials at different points during treatment and specifically in case of relapse or end-stage cancer and supporting negotiation processes during informed consent conferences results in enhanced comprehension[67,73].

The ethical permissibility of phase I clinical trials in pediatric oncology can be based also on parents' and patients' accounts that when properly informed, they cherish contributing to help others facing a cancer diagnosis[64,75]. Moreover, the involvement not only of parents, but of children is central to treating them as ends in themselves[27] and in turn requires proper communication to uncover and respect personal values in decision-making[71]. Discussions about research participation at the end-of-life have to extend to incorporate sharing of information that is important to patients also as a requirement for obtaining valid assent[76]. When patients' abilities are not sufficient to allow participation in decision-making, still it is important to acknowledge the contribution that children are making through their bodies, as part of their whole person[71]. This recognition can provide an ethical justification for non-beneficial research with children lacking capacity and has to be supplemented by good physician-parent communication at the end-of-life[70]. Parental understanding of the purpose of phase I clinical trials and of the altruistic aspect of their consent can contribute to recognizing children as body-subjects and help patients express choices at the end-of-life[70,71,77].

Communication about phase I and end-of-life care

Besides phase I clinical trial participation, families and patients are faced with other difficult options - treatment withdrawal or withholding, transition to terminal care[70,78]. It is essential to

present and discuss all these care options when talking about non-beneficial research, so that parents and patients understand their choices and also they are able to separate altruistic participation from that with therapeutic chances[71,79].

Guidelines that promote the provision of better palliative care emphasize the importance of eliciting patients' wishes[80,81]. However, patients' voices are rarely given a place in decision-making at the end-of-life[82]. A recent study in pediatric oncology highlights the aggressive care most teenagers receive in the last months of life[78,83]. The authors hypothesize that in cases of poor prognosis and when cure is no longer an option patients are rarely involved in treatment planning and discussions about limiting treatment[78,83]. This robs families and patients of chances to say goodbye and does not support the provision of high quality care that respects personal and cultural beliefs at the end-of-life[84-86]. The incremental involvement of children for which treatment failed is aligned with their best interest. At this stage, their views should guide all decisions to be made, as there is no risk of unwise decisions to their health[84]. Honoring their wishes for a good death[84], to discuss death and dying and leave memories for families and friends is a determinant of best care. It helps empower children in their most vulnerable state by giving them more latitude to decide. This would mean to respect them as people and tend to their voices and fears when cure is not possible[39,41,82].

Research shows that communication at the end-of-life is particularly difficult in view of families' attitudes. Parents often exhibit reticence in discussing passing away with patients and physician support for families is insufficient[70,77,87]. Communication and physician sharing of information to prepare parents for the challenges of care in the last stages of the illness is inadequate and leaves parents doubting what is best for their child. Parents' needs are centered on doing the right thing and "being a good parent"[70,88], making communication the most important indicator of providing good care. Physician actions in this phase are not equally oriented toward relational components of care[88], but it is their duty to help parents

transition from big to small hopes. Such actions do not imply a destruction of hope, but focus on helping parents clarify their expectations - from hoping to see their children cured to aiming for more good quality time with their children and meaningful goodbye[30,58,77].

Culture and communication

Differences regarding communication for children diagnosed with cancer have been attributed to cultural paradigms[5,13]. The influence of social norms and the adoption of a particular belief system is most obvious in regard to disclosure of diagnosis practices[47,89,90]. On one hand, professional behavior in the medical setting in one country is be commanded by a primordial respect for autonomy, while in another country is driven by beneficence and relational autonomy[40,91,92]. The latter two principles award physicians more discretion in judging what is harmful for relatives and patients[11,47,93]. On the other hand, disclosure may be determined by broader cultural norms and values within society and those held by families. Views on cancer and fatalism, truth telling and bearing responsibility for protecting loved ones and perceptions regarding hierarchical relationships with physicians influence behavior in communicating about cancer[46,91,94,95]. Given these variations, physician responsibilities toward informing the child patient appear to be subjected to geographical or cultural interpretations[47,96]. Solutions for harmonizing patient right to information with perceived cultural attitudes that go against full and open communication are ethically complex[25,40,46,56]. Attributing a divergence of opinions on disclosure to children to cultural backgrounds runs the risk of assessing the situation as an unsurmountable clash between professionals' and parents' positions. It easily results in labelling parents who oppose (full) disclosure as difficult families and patient cases, feeding into cultural biases[97,98]. These may be potential

sources of conflict or break-down in communication, when parties do not share the same values[40,46,47].

As the studies conducted in pediatric oncology centers in Romania and Switzerland show, physicians in the two countries have different approaches toward disclosure. Swiss oncologists are sternly in favor of delivering the cancer diagnosis to patients and inform parents about this approach matter-of-fact[41]. In Romania, physicians are more cautious regarding parental hesitation and wishes[40,56]. Oncologists share a belief that children should be informed, but manifest prudence in relation to the probability that patients, especially adolescents, may become depressed[32,56,99], and leave significant leeway for parents' preferences[40,56].

Despite the country differences observed in regard to frequency of requests to withhold the diagnosis from patients and physicians honoring them, variable behaviors were spotted in both settings. Disclosure of diagnosis takes different forms and families born or acculturated to the same environment hold different views on if and how, as well as when to inform children about the diagnosis[40,41,56]. In Switzerland, where there is a usual parental agreement to share information with children, few parents, at least initially, did not conform to the status quo on disclosure[41]. Likewise, in the Romanian sample parental approaches to communicating the diagnosis were diverse, although most parents' default reaction to informing children was protecting them from the truth, as much as possible[40]. In spite of this prevailing stance, some parents who at first believed that it would be best not to share information with children changed their mind during the day one talk or soon afterwards[40,56]. These accounts show that communication regarding difficult news is not strictly subjected to perceived cultural norms and reveal that parents' inclination toward non-disclosure can be successfully challenged[40,46,56]. In fact, preferences for not communicating the diagnosis can be attributed to what families are used to in terms of directly informing children and

customary family structures and values[40,41,56]. Expectations in the delivery of care also play a role in forming preferences to limiting information, but can be shifted by physicians, including during the first talk[20,40,46,56].

At the same time, physicians associated with or practicing in a particular culture may not always hold a firm attitude toward open disclosure. In an open system where the standard of care is to tell patients and families all facts, they are more at ease when disclosing a life-threatening disease. Such approach can easily fall under a medicocentric view, focused primarily on physiological aspects of illness[100] when physicians while breaking bad news also initiate a "project of cure"[25]. This seemed to be the case for disclosure of diagnosis in Swiss centers[41,42]. However, even when practicing an open-model of disclosure physicians find it difficult to discuss poor prognosis, terminal illness and lack of standard treatment[11,29,101]. For particular patient circumstances and contexts some physicians choose to limit information provision at diagnosis in what concerns significant uncertainty in achieving a cure[39,41]. Participant physicians in Switzerland sometimes displayed such tendencies to avoid burdening patients with negative and excessive information about the illness[41].

Professional guidelines, despite advocating for an open and full communication about cancer also mention that disclosure is not a one-time event[24,25]. They leave sufficient room for interpretation and it is important to distinguish what form of disclosure is best suited for a particular child and her parents, especially on the background of ethnically diverse patient populations[43,46,91] . Classifying parents into binary categories - pro-disclosure and non-disclosure - ignores the continuum on which sharing of information regarding diagnosis takes place and is grounded more on assumptions that behavior and wishes are attributed to a certain culture[40,41,43].

Physicians who engage in the endeavor of assessing what is preferable and adequate for individual cases should rely on strategies of exploration of parents' and subsequently

patients' attitudes. How one thinks about a topic is based on individual experiences, the perceived situation of others or their reports regarding that subject, personal beliefs and moral views. Parents collect their information from different sources, such as the Internet, newspapers, including lay persons in their community, circle of friends[39,40,102]. Attitudes incorporate cultural elements, popular and sometimes religious ideas on the topic[103], but go beyond assumptions that individuals from a predominant biomedical culture have to approach cancer strictly from a cognitive and factual perspective[104,105] . At the same time, the attitudes a person has in relation to cancer drive behaviors that are independent of rational thoughts and educated opinions[106].

An explanatory model of illness for communication in pediatric oncology (EMICPO)

In order to grasp the attitudes that parents and children have regarding cancer and medical care physicians can employ a model of explanatory illness[107,108]. The basis for the model is physician's acknowledgment of the fact that parents and children may have specific or developing attitudes in relation to the patient's health condition, even before the cancer diagnosis is established[108]. Clinicians recognize and accept "different versions of the clinical reality" that are deserving of equal consideration and respect[108]. Oncologists have to also consider that these attitudes trigger individual emotional responses to disclosure of a cancer diagnosis and influence preferences for initial communication with the child patient[20,32,40,56]. Attitudes are fundamental elements for parents' and patients' first gut feeling reaction to diagnosis and can extend into communication about the illness and their behavior during treatment[56,107]. For example, in the Romanian sample many parents purposefully avoided the word cancer as a way of obscuring the potentially life-threatening nature of the illness. Both parents and physicians believed that the connotation between cancer and dying is always

present and did not want patients to think about this likelihood[40,56]. In Switzerland parents and physicians would name the disease but also mention that cancer in children is different than in adults and that treatments are better[41,43]. This is suggestive of efforts to instill and emphasize confidence in the efficacy of treatments in curing cancer and provide hope[25,109]. However, even in Swiss centers communication at diagnosis regarding risk factors and difficulties in treating rare cancers in children becomes more ambiguous in cases of poor prognosis. This perhaps is due to a belief that addressing the dangerousness of some malignancies would likely lead parents and patients to question the chances of surviving the illness[41,42].

At the same time, physicians need to reflect on their own attitudes toward cancer, which are shaped by their medical training and belonging to a subculture centered on understanding physiological manifestations of disease rather than patients' experiences of illness[108,110,111]. Embracing either the patient's view of illness or the physician's isolated image of cancer as disease is insufficient to gain a comprehensive understanding that educates care practices[108]. Apart from acknowledgment that (different) attitudes, of parents, physicians and patients exist, an explanatory model of illness requires physicians to engage with these attitudes. First, oncologists have to elicit parents' views, beliefs and insecurities about the child's illness. Kleinman's model offers a guiding set of questions which physicians should make use of during the first encounter with parents[108]. This covers several domains of parental thinking regarding: cause of child feeling sick and time of occurrence, manifestation of the problem, its severity and expectations of what treatment is adequate[108]. Annex 1 presents a developed model of questions, EMICPO, based on Kleinman's framework[108] and drawing from the SPIKE tool for communicating bad news[54] (Please see Annex 1). Processes in EMICPO focus on what parents worry about regarding the diagnosis, how they believe children should be told, when and by whom, in relation to prior experiences with cancer. The EMICPO can

be adapted to elicit what children know so far, what they believe is the problem, what would they like to know more and from whom, and who should be present during discussions (Annex 1). Questions can be addressed in one or several appointments, based on parents' and children's wishes and their emotional state[25,40].

Second, physicians need to address these attitudes, by offering tailored information to what they previously heard and by correcting misunderstandings. This process, of "talk-tell-talk"[15,54], helps in the clinical practice to educate parents by comprehensively taking into account their experiences with the illness while identifying individual levels of adaptation of medical information to lay terminology. EMICPO is a perpetual comparison between physicians' and parents' attitudes based on information sharing. The model thus recognizes and blends parental with physician views and negotiates a common model of understanding of illness and experiences with cancer[108]. It is a mixed model of physiological and psycho-social explanations of cancer acceptable to both parties[110] (Annex 1). The EMICPO incorporates medical facts, including the reality of an ever present uncertainty, along with personal stories of relatives who did not survive cancer and illness associated fears. At all times, physicians must reassure parents that they may ask questions at any time and that expressing their thoughts, beliefs and needs is welcomed by and helpful for physicians, as well as for care[40,41,43,46,56,108].

The use of EMICPO

In pediatric oncology moral conflicts and ethical dilemmas are often the result of postponed discussions about sensitive issues of care, divergent views regarding treatment course and care goals, and parent-physician tensions[74,101,112]. Lack of adequate communication between families and clinicians may develop into latent conflicts that threaten fiduciary responsibilities of both parents and physicians when providing care for patients. This situation may escalate to the point that external consultation with an ethics committee is

required[74,112]. The use of the EMICPO is a helpful tool to use throughout parent-physician encounters, a reiterative operation in providing care and possibly diminishes risks of conflict. In both the Romanian and the Swiss clinics the use of EMICPO would have showed that full disclosure in one meeting is not always well received by families[40,42,43]. It could have highlighted parents' needs for clarifying information provided for decision-making about treatment, research participation and fertility preservation, and stirred physician efforts in the right direction[39,41,43].

Clinicians can adapt questions from the EMICPO to specific context and use it in communication with parents and, when adequate, patients during rounds and routine check-ups. It serves also a control for past information communication after parents had time to absorb it and facilitates clarification of things that were left unclear and ask further questions[43]. Especially when new information is presented regarding treatment outcomes, medical tests and risk factors, clinicians should assess parents' and patients' attitudes. Continuing to elicit their views and attitudes regarding their experience facilitates identifying their preferred and changing roles in medical communication and decision-making[39,41,56]. Therefore a model of exploration is necessary to allow for identification of what choices can be made collaboratively between parent-patient-physicians, which between parent-physician and which should be offered to patients[39,57]. This can positively affect satisfaction with the provided care.

If in communicating with families physicians start with an explanatory model of illness and always support and try to help parents and patients to verbalize and explore their fears, families will be better prepared. EMICPO can particularly facilitate communication in case of difficult decision-making about participation in RCTS, relapse or when facing the end-of-life. It therefore makes difficult talks and decisions around treatment failure, prognosis and dying easier. Parents who are continuously supported in dealing with and thinking about what is

important can better frame their values. The EMICPO can be used in relation to hope management and getting parents to talk about and focus on small hopes (e.g. maintaining an acceptable quality of life, returning the child to school, attend graduation, spending more time with family, reduce the number of hospital admissions). Furthermore, this exploratory model of communication in pediatric oncology is likely to better answer to diverse family beliefs and improve care for ethnic groups[40,56].

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Implications for practice and future research

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Results of this doctoral research and the above analysis have direct implications for clinical practice and emphasize several aspects that need to be investigated further:

- Disclosure of diagnosis is a complex process influenced by different individual or cultural factors that need to be acknowledged by clinicians.
- Communication in pediatric oncology is deficient in several areas neglected during routine medical encounters which result in mundane harms.
- There is a need to develop, assess and implement disclosure and communication protocols with parents and children that take into account explanations of illness.
- Physicians have a duty to care also for parents and this requires identifying if parents' reactions to diagnosis have a pathological component and take appropriate measures.
- Conduct regular assessment of information understanding and needs for both parents and children, and address them in order to improve decision-making.
- Research has to investigate patients' perspective in case of non-disclosure and identify risks.
- Test and further refine a modified explanatory model of illness, the EMICPO; assess its effects on communication about diagnosis, prognosis, treatment, research and end-of-life decision-making.
- Explore what parents and when adequate patients want to know about short- and long side-effects and risks and discuss how they would deal with them. Additionally, physicians need to address the issue of different sources of information.

- Current standards of decision-making and efforts of distancing from paternalistic practices in decision-making pose difficulties for some parents and undercut the importance of relational autonomy.
- Place greater emphasis on elements of micro-ethics, such as building relationships, which can be more valuable for parents.

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Conclusions

Conclusions

Cancer's burdens cannot be disentangled from families' experiences with pediatric oncology care, even in case of prolonged remission. There are different subtle ways in which parents and children are harmed and the relationships between them and physicians tested. Communication in pediatric cancer care poses challenges that go beyond medical information and is a central aspect of providing high-quality care. Clinicians and families in different settings can face various difficulties in communicating about the illness, treatment and options. Culture could account for some of these differences and provide different ethical solutions. However, as the studies in Switzerland and Romania reveal there is a need to explore parental views and attitudes by looking at more than culture. The meaning of illness, emotional turmoil, and previous experiences with cancer and encounters with clinicians serve better to define differences in communicating about and disclosing cancer diagnosis. The EMICPO creates opportunities to take an active approach to elicit and share information with parents and patients. It can be applied to different areas and by its reiterative feature facilitates communication at the time of diagnosis, during treatment and decision-making processes, including regarding therapeutic and non-beneficial research and end-of-life. Its use and the results it has on communication need to be tested in practice.

Annex 1

Annex 1

Table I. Explanatory model of illness for communication in pediatric oncology (EMICPO)*

Questions eliciting attitudes at diagnosis		
To be explored with parents		**To be explored with patients**
Assessing the situation and understanding	What do you think is the matter with your child?	Questions directed at parents can be adapted for minor patients, according to level of understanding. Physicians may skip questions that they consider inadequate.
	What do you think is causing this problem to your child?	
	How do you think this problem started?	
	When did it start? Why do you think it started then?	
	What do you think is this sickness doing to your child?	
	What do you think about the severity of your child's situation?	
	How worried are you? About what do you worry/ about what do you worry most?	
	What have you been told so far about your child's condition?	
Assessing attitudes toward disclosure of diagnosis	What have you discussed with your child about his sickness so far?	What do you want to know about your sickness?
	What do you think your child thinks about his sickness?	
	How do you think your child should be told about the illness?	
	What do you think your child should be told? When should he be told? Who do you think should talk to your child?	
	What worries you most about talking to your child about this illness?	

	Is there something that you find difficult to share with your child? Why do you think that is?	
	How do you think your child will react to being told the diagnosis?	
Eliciting sources of information and needs	Did you talk with somebody else about your child's sickness? With whom (family, friends, child's teacher)? What did they say?	Questions should be adapted for individual patients, as judged to be appropriate.
	Did you get information from other sources (Internet, news, movies)?	
	What do you think about what others said to you? What do you think about what you learnt from other sources?	
	What questions do you have so far? What else do you think you need to know?	What questions would you like to ask me?
		Who do you want to talk to you about your sickness?
Assessing what kind of care was sought so far and what are the expectations for future care	What was done so far for your child's sickness?	Questions should be adapted for individual patients, as judged to be appropriate. Questions may be skipped.
	What is your understanding of the reasons for the medical tests done so far?	
	What do you think about the kind of treatment your son should receive?	
	What do you hope that the treatment will do?	

[*] The design of the EMICPO model is based on Kleinman A, Eisenberg L, Good B. Culture, illness, and care: clinical lessons from anthropologic and cross-cultural research. Annals of Internal Medicine. 1978;88(2):251-258, and an adaptation of the SPIKE tool for communicating bad news conceptualized by Baile WF, Buckman R, Lenzi R, Glober G, Beale EA, Kudelka AP. SPIKES—A Six-Step Protocol for Delivering Bad News: Application to the Patient with Cancer. The Oncologist. 2000;5(4):302-311.

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Appendices

Appendices

Study Title

Original Study Title in English: **Competency and autonomy of children and adolescents in pediatric oncology decision making**

Study Title in Romanian: **Competenţa şi autonomia minorilor şi adolescenţilor în procesul decizional în oncologia pediatrică**

Interview Guide for Parents - Romanian study

English version

Dear participant: Thank you for agreeing to take part in this study and for sharing your information with us. You are being asked to take part in the research study of "**Competency and Autonomy of Children and Adolescents in Pediatric Oncology Decision Making**" because your child was diagnosed with cancer and/or is currently undergoing cancer treatment in a Romanian Oncology Centre. We understand that this is a very sensitive issue and we would seek to be as considerate as possible. If you feel uncomfortable responding to any question, please communicate this to me.

During this interview session, we would like to learn about the condition of your child and the quality, content, and process of decision making regarding his/her illness. The information that we gain from you will help us understand decision making processes and contribute towards debates concerning a minors' decision making autonomy concerning

health care. After the interview, we will request you to complete a very short 1 page survey. Please be assured that your responses and all the information/data you provide are completely confidential and anonymous. We thank you for agreeing to be part of the study and answering the following questions.

***Note to the interviewer: For questions marked with *, customize the question based on the context.**

For example, for question 1.e, if prognosis is poor - ask questions e.1., e.2.; if prognosis is not poor, continue with question 1.f.

1. **Understanding the situation:**
   a. Let me start by asking you about your family. Could you tell me about them?
   b. Please briefly describe to me the circumstances regarding your child's cancer diagnosis?
   c. How did you first come to believe that your child might have cancer?
   d. When was the diagnosis made? How old is your child now?
   e. What was the prognosis the physician gave you?*

      ***(Note to the Interviewer: Ask the following two questions only if the parent described the chances as poor)**

      1. When did you understand that the chances of cure for your child are rather low?
      2. Could you please tell me more about how you see the chances of cure for your child?

   f. Was your child present during the diagnosis discussion? Why or why not?*

   ***(Note: If child present, ask question g, else skip to h.)**

g. How did you feel about your child's presence in regards to the diagnosis communication?

h. Since the diagnosis was made, have you informed your child about the diagnosis?

i. How did you do so? Could you tell me the words you used? For example: what was the first sentence you used to start the discussion? Why did you choose the words you chose?

j. Did you discuss the prognosis with your child? Did you explain him/her the chances of cure?

k. Who initiated this discussion? How did you feel during the discussion?

2. Treatment options, discussions, and decision making

a. What were the treatment options?

b. Who was present when treatment options were discussed?

c. How were these treatment options described to you and/or your child by your physician?

d. Were these options clearly and carefully explained to you and/or your child by your physician?

e. If your child was not present during treatment discussions, have you discussed treatment or non-treatment options with your child?*

***(Note: if answer is no, skip to question g.)**

f. What did/do you think about this discussion with your child?

g. What treatment options were selected? And why?

h. How were decisions made about your child's illness and treatment choices?

i. Who was involved in this decision making process?

j. To what extend was the child involved? Please provide reasons for extent of involvement or non-involvement.

k. How comfortable were you with the manner and content of these discussions?

l. Were you satisfied with these discussions?

m. Please explain what factors contributed to greater or lower satisfaction.

n. Would you change anything retrospectively? e.g. Is there anything in your opinion that would have made these discussions more beneficial?

3. **Inclusion or Exclusion of child in discussion:** The following questions relate to the capacity of children to make decisions concerning their health and medical treatment, in particular, if they are severely ill. In society there is a debate about whether and how much children or minors should be involved and given the right to make decisions. We, therefore, would like to gather your opinion on this topic.

a. In your previous response, you stated that your child was present (absent) during treatment discussions. Could you explain the reasons for this?

b. Why do you think it was necessary/appropriate to include (exclude) your child from these treatment decisions?

c. Under what conditions would you absolutely include (exclude) your child in such discussions? Please explain these conditions.

d. Generally, when parents include (exclude) children from making such decisions, what kind of reasons do you think, for example values and attitudes, are associated with it?

e. In your opinion, should children in general, be allowed to make health care related decisions? Please explain your perspective.

- Would your response be the same or differ based on the following:

- Age of the child: younger than 8 years or older than 8 years (or another age limit?)
- Type and seriousness of the disease (depending on curative chances/prognosis)
- Behavior of the child
- Your religion or personal beliefs

**********Thank You **********

Dear Participant: We thank you for taking the time to complete the following.

A. Demographic Information:

a. Age: _____ Year of Birth: _____

b. Sex: ☐ Male ☐ Female

c. Nationality: ☐ Romanian ☐ Other – _____

Country of origin: _____

d. Languages: ☐ Romanian ☐ Hungarian ☐ German

☐ English ☐ French

☐ Other (specify): _____

e. Marital Status: ☐ Married ☐ Divorced ☐ Widowed

☐ Never Married ☐ Other – Specify _____

f. Religion: ☐ Orthodox ☐ Catholic ☐ Protestant

☐ Other (specify): _____

g. Education: ☐ No qualification ☐ Vocational ☐ High School

☐ College Degree ☐ University/Graduate Education

h. Number of children: _____

**********Thank You **********

Titlul Proiectului:

Titlul original în engleză**: Competency and autonomy of Children and Adolescents in pediatric oncology decision making**

Titlul în română: **Competenţa şi autonomia minorilor şi adolescenţilor în procesul decizional în oncologia pediatrică**

Ghid interviu pentru părinţi - Romanian study

Romanian version

Stimat/ă participant/ă: Vă mulţumim pentru că aţi acceptat să participaţi la acest studiu şi să ne împărtăşiţi informaţii în legătură cu dumneavoastră. Vi s-a cerut să participaţi la studiul de cercetare "**Competenţa şi autonomia minorilor şi adolescenţilor în procesul decizional în oncologia pediatrică"** deoarece copilul dumneavoastră suferă de cancer sau/şi în prezent primeşte tratament pentru cancer în cadrul unui spital din România. Suntem conştienţi de caracterul sensibil al studiului de cercetare şi de aceea vă asigurăm că vom trata cu cel mai mare respect acest subiect. Dacă o întrebare vă jenează, vă rog să îmi spuneţi.

În cursul acestui interviu, încercăm să cunoaştem starea actuală de sănătate a copilului dumneavoastră, precum şi continutul, modul de derulare al procesului de luare a deciziilor medicale cu privire la boala sa. Informaţiile pe care ni le veţi furniza ne vor ajuta să înţelegem mai bine procesul de luare a deciziilor, ce va sta la baza dezbaterilor pe tema autonomiei, respectiv a recunoaşterii dreptului minorilor de a lua decizii cu privire la starea lor de sănătate şi a respectării acestui drept. După ce interviul se va incheia, vă vom ruga să completaţi un scurt chestionar de o pagină. Vă asigurăm că atât răspunsurile dumneavoastră, cât şi informaţiile/datele pe care ni le veţi oferi vor fi anonimizate (nu vom folosi numele

dumneavoastră sau al copilului dumneavoastră sau alte date care v-ar putea identifica). Toate informaţiile/datele vor fi tratate de manieră strict confidenţială. Vă mulţumim pentru că aţi acceptat să luaţi parte la acest studiu şi să ne răspundeţi la întrebări.

***Notă personală destinată persoanei ce va conduce interviul: în ceea ce priveşte întrebarile marcate cu *, avem rugămintea de a le adapta în funcţie de răspunsurile oferite anterior. Spre exemplu, în ceea ce priveşte întrebarea 1.d., dacă răspunsul este pesimist/negativ/prost, treceţi la întrebarea 1.e.; dacă răspunsul este foarte bun/bun etc., vă rugăm să adresaţi întrebările 1.d. 1 şi 1.d.2..**

1. Înţelegerea situaţiei:

a. Aş dori să încep prin a vă pune o întrebare cu privire la familia dumneavoastră. Îmi puteţi spune mai multe despre familie şi membrii ei?
b. Îmi puteţi descrie pe scurt în ce circumstanţe copilul dumneavoastră a fost diagnosticat cu această boală ?
c. Când a fost diagnosticat copilul dumneavoastră? Ce vârstă are în acest moment?
d. Care a fost prognosticul oferit de medici?

***(Notă personală adresată persoanei care va conduce interviul: NU adresaţi următoarele două întrebări decât dacă părintele a răspuns pozitiv confirmând că prognosticul este unul pesimist/negativ.)**

1. Când aţi înţeles că posibilitatea unei vindecări pentru copilul dumneavoastră e puţin probabilă?

2. Îmi puteţi spune mai multe despre cum vedeţi şansele de vindecare/ameliorare a copilului dumneavoastră? Cum v-aţi simţit când vi s-a comunicat diagnosticul ? Şi cum vă simţiţi acum?

e. Copilul dumneavoastră a fost prezent în timpul discuţiilor cu privire la dignostic? De ce DA? Sau de ce NU?*

***(Notă: Dacă acesta a fost prezent, a se continua cu întrebarea g. Dacă NU, a se trece direct la intrebarea h.)**

f. Ce părere aveţi şi ce aţi simţit în legătură cu prezenţa copilului dumneavoastră în cursul comunicării diagnosticului?

g. După stabilirea diagnosticului, ce i-aţi spus copilului dumneavoastră cu privire la diagnostic?*

***(Notă în cazul în care părintele nu a discutat diagnosticul cu copilul se adaugă intrebarea: De ce Nu?)**

h. Cum i-aţi comunicat diagnosticul? Aţi putea să descrieţi pe scurt ce cuvinte/limbaj aţi folosit? Spre exemplu, cu ce propoziţie aţi deschis discuţia? De ce aţi ales aceste cuvinte?

i. Aţi discutat despre prognostic cu copilul dumneavoastră? I-aţi vorbit despre şansele de vindecare?

j. Cine a iniţiat această discuţie?

k. Cum v-aţi simţit dumneavoastră în timpul acestei discuţii?

l. Dacă vă gândiţi la acel moment, credeţi că a fost bine, mai bine să discutaţi deschis despre aceste probleme cu copilul dumneavoastră? Vă rog să explicaţi.

2. Opţiuni terapeutice, discuţii şi luarea deciziilor

a. Care au fost opţiunile terapeutice pe care vi le-au dat doctorii?

b. Cine a fost prezent în cadrul discuţiilor referitoare la opţiunile de tratament?

c. Cum au fost pentru dumneavoastră şi/sau pentru copilul dumneavoastră descrise de către medic aceste opţiuni de tratament?

d. Credeţi că aceste opţiuni de tratament v-au fost explicate clar şi pe îndelete dumneavoastră şi/sau copilului dumneavoastră de către medic?

e. Dacă copilul dumneavoastră nu a fost prezent în timpul acestor discuţii, i-aţi vorbit depre opţiunile de tratament sau despre posibilitatea de a nu accepta tratamentul medical?*

***(Notă: dacă răspunsul este NU, treceţi la g.)**

f. Ce aţi crezut/ Ce credeţi despre discuţia cu copilul dumneavoastră?

g. Ce opţiuni de tratament medical au fost alese în final? De ce aţi ales aceste opţiuni?

h. Cum au fost luate deciziile legate de boala diagnosticată şi cu privire la opţiunile de tratament pentru copilul dumneavoastră?

i. Cine a a luat parte în procesul de luare al acestor decizii?

j. În ce măsură a fost implicat copilul dumneavoastră în acest proces?

k. Doriţi, vă rog, să explicaţi care sunt motivele pentru care implicarea copilului dumneavoastră a avut loc în mai mică sau mai mare măsură sau deloc?

l. Cât de mulţumit/ă sau liniştit/ă vă simţiţi în ceea ce priveşte modul în care s-au purtat aceste dicutii şi conţinutul lor?

m. Sunteţi mulţumit/ă de rezultatul acestor discuţii?

n. Vă rog să explicaţi care sunt elementele cele mai importante şi cele mai puţin importante care au contribuit la gradul de mulţumire/nemulţumire cu privire la aceste discuţii.

o. În retrospectivă, aţi dori să schimbaţi ceva referitor la aceste discuţii? Spre exemplu, există elemente care au lipsit în cadrul discuţiilor, dar care potrivit dumneavoastră ar fi fost binevenite?

3. Includerea sau excluderea copiilor/adolescenţilor din cadrul discuţiei cu privire la tratament: Întrebările următoare se referă la capacitatea şi dorinţa exprimată a copiilor/adolescenţilor de a lua decizii cu privire la starea lor de sănătate şi tratament, în mod special în cazul bolilor grave. În prezent există o dezbatere în mediul medical şi public cu privire la implicarea copiilor şi/sau adolescenţilor în astfel de discuţii şi în ce măsură aceştia au capacitatea şi posibilitatea de a participa la luarea deciziilor referitoare la starea lor de sănătate. Din aceste motive, ne interesează punctul dumneavoastră de vedere pe această temă.

a. În răspunsurile anterioare mi-aţi spus că în cadrul discuţiilor legate de tratament copilul dumneavoastră a fost prezent (absent). Îmi puteţi explica motivele pentru aceasta?

b. De ce credeţi că a fost necesar sau potrivit să îl implicaţi (excludeţi) pe copilul dumneavoastră din cadrul acestor discuţii?

c. În ce condiţii v-aţi implica (exclude) copilul din asftel de discuţii? Doriţi vă rog să precizaţi aceste condiţii mai detaliat?

d. În general, când părinţii îşi implică (exclud) copiii din astfel de discuţii, care credeţi că sunt motivele pentru astfel de atitudini (aceste decizii)? De exemplu, ce criterii, valori sunt luate în considerare?

e. După părerea dumneavoastră copiii, în general, ar trebui să aibă dreptul de a participa la luarea unor decizii ce privesc starea lor de sănătate? Vă rog să explicaţi mai detaliat răspunsul dumneavoastră:

- Răspunsul dumneavoastră ar fi acelaşi sau diferit în funcţie de:
    - Vârsta copilului: dacă are mai puţin sau mai mult de 8 ani (sau o altă limită a vârstei)?
    - Tipul şi gravitatea condiţiei medicale: (în funcţie de şansele de vindecare)
    - Comportamentul copilului
    - Religia dumneavoastră sau convingerile personale

**********Mulţumesc **********

Stimat/ă participant/ă: Vă mulţumim în avans pentru dorinţa de a completa cheastionarul următor:

B. Informaţii demografice pentru părinţi:

a. Vârsta: _____ Anul naşterii: _____

b. Sex: ☐ Masculin ☐ Feminin

c. Naţionalitate: ☐ Română ☐ Alta – _____

Ţara de origine: _____

d. Limbi vorbite: ☐ Româna ☐ Maghiara ☐ Germana

☐ Franceza ☐ Engleza

☐ Alta (precizaţi): _____

e. Starea civilă: ☐ Căsătorit/ă ☐ Divorţat/ă ☐ Văduv/ă

☐ Necăsătorit/ă ☐ Alta- Precizaţi _____

f. Religia: ☐ Ortodoxă ☐ Catolică ☐ Protentantă

☐ Alta-Precizaţi _____

g. Educaţie/ Studii: ☐ nicio calificare ☐ şcoală primară

☐ Gimnaziu ☐ Studii liceale

☐ şcoală profesională ☐ Bacalaureat

☐ Diplomă de absolvire a unei facultăţi/ studii superioare

h. Numărul de copii: _____

**********Mulţumesc **********

Study Title

Original Study Title in English: **Competency and autonomy of children and adolescents in pediatric oncology decision making**

Study Title in Romanian: **Competenţa şi autonomia minorilor şi adolescenţilor în procesul decizional în oncologia pediatrică**

Interview Guide for Physicians - Romanian study

English version

Dear participant: Thank you for taking the time and agreeing to participate in this study. You are being asked to take part in the research study of "**Competency and Autonomy of Children and Adolescents in Pediatric Oncology Decision Making**" because you are a treating physician in a Romanian Oncology Centre for children diagnosed with cancer or who are currently undergoing cancer treatment. The information that we gain from you will help us understand decision making processes and contribute towards debates concerning a minors' decision making autonomy concerning health care.

During this interview session, we would like to learn about the condition of your patient, the quality, content, and process of decision making, and the patient's competency. After the interview, we will request you to complete a very short 1 page survey. Please be assured that your responses and all the information/data you provide are completely confidential and anonymous. We thank you for agreeing to be part of the study and answering the following questions.

***Note to the interviewer: For questions marked with *, customize the question based on the context.**

For example, for question 2.c., if information is provided together - ask that question; if information is discussed separately, rephrase the question for parents and child separately.

1. **General information about the patient's case**
   a. How long have you known the patient and his or her family?
   b. When did you first see the patient and make the diagnosis?
   c. How old is the patient now?
   d. What were the diagnosis and prognosis?
   e. Who did you disclose the diagnosis and prognosis to? If your patient was not present, please indicate why.
   f. What was the parent's reaction when the diagnosis and prognosis were made?
   g. If you discussed the diagnosis with the child, what was the child's reaction?
   h. If you discussed the prognosis with the child, did you explain him/her the chances of getting better?
   i. If you did not discuss the prognosis with the child, why not?
   j. Who initiated this discussion related to diagnosis and prognosis with the parents and/or the child? How did you feel during the discussion?
   k. Can you explain a little more the circumstances and content of these discussions?

2. **Treatment options, discussions, and decision making**
   a. What were the treatment options?
   b. Who did you discuss the treatment options with?

c. How did you describe the treatment options to the child and/or the parents?*

d. How comfortable did you feel discussing treatment options? Please explain.

e. Are alternative treatments available for this case? Please explain.

f. What were the clinically reasonable choices?

g. Did you discuss the pros and cons of the choices? Why and why not?

h. In your opinion, how well did the child and/or the parent understand the treatment options you provided them with?*

i. Were these discussions carried out with the child and/or the parent? Please specify with whom (if child excluded, explain why).

j. What treatment options were selected?

k. Who was involved in the decision making process?

l. How were decisions made about the child's illness and treatments? (e.g., what were the important issues discussed, how were the opinions of those involved heard, and how was consensus formed.)

m. To what extend was the child involved? Please provide reasons for extending involvement or non-involvement.

n. How engaged were you in these decisions?

o. Were your opinions and expertise invited? How comfortable did you feel with that?

p. Were you satisfied with these discussions?

q. Please explain what factors contributed to greater or lower satisfaction.

r. Would you change anything retrospectively? e.g. Is there anything in your opinion that would have made these discussions more beneficial?

3. Inclusion or Exclusion of child in discussion: The following questions relate to the capacity of children to make decisions on behalf of their health and medical treatment, in

particular if they are severely ill. In society there is a debate about whether and how much children or minors should be involved and given the right to make decisions. We, therefore, would like to gather your opinion on this topic.

a. In your previous response, you stated that the child, i.e., patient, was present (absent) during treatment discussions. Could you explain the reasons for this?

b. Why do you think it was necessary/appropriate to include (exclude) the patient from these treatment decisions?

c. Under what conditions would you absolutely include (exclude) patients in such discussions? Please explain these conditions.

d. Generally, when parents include (exclude) their child from making or taking part in such decisions, what kind of reasons, for example values and attitudes, do they give you?

e. In your opinion, should this child be included in decision making. Explain why or why not.

f. In your opinion, what role should the parents play in making decision?

g. In your opinion, should children in general, be allowed to make health care related decisions? Please explain your perspective.

- Would your response be the same or different based on the following:
    - Age of the child: younger than 8 years or older than 8 years (or another age limit?)
    - Type and seriousness of the disease: terminal or non-terminal
    - Behavior of the child
    - Your religion or personal or professional beliefs

**********THANK YOU**********

Dear Physician: We thank you for taking the time to complete the following.

Physician demographic Information:

f. Age: _____ Year of Birth: _____

g. Sex: ☐ Male ☐ Female

h. Nationality: ☐ Romanian ☐ Other – _____

Country of origin: _____

i. Languages spoken: ☐ Romanian ☐ Hungarian

☐ German ☐ English

☐ French

☐ Other (specify): _____

j. Marital Status: ☐ Married ☐ Divorced ☐ Widowed

☐ Never Married

☐ Other – Specify _____

k. Medical Specialty: _____

l. Years of experience in pediatric oncology:

☐ 0 – 4 years ☐ 5 – 8 years

☐ 9 – 12 years ☐ More than 12 years

m. Religion: ☐ Orthodox ☐Catholic ☐ Protestant

☐ Other (specify): _____

**********THANK YOU**********

Titlul Proiectului:

Titlul original în engleză**: Competency and autonomy of children and adolescents in pediatric oncology decision making**

Titlul în română: **Competenţa şi autonomia minorilor şi adolescenţilor în procesul decizional în oncologia pediatrică**

Ghid interviu pentru medicii participanti la proiect - Romanian study

Romanian version

Stimat/ă participant/ă: Vă mulţumim pentru că aţi acceptat să alocaţi din timpul dumneavoastră şi să participaţi la acest studiu. Aţi fost invitat/ă să participaţi la proiectul de cercetare denumit: "**Procesul decizional la finalul vieţii: Competenţa şi autonomia minorilor şi adolescenţilor în oncologia pediatrică"** deoarece sunteţi doctor specialist în cadrul unui spital universitar, clincă oncologică sau insitut oncologic din România, şi aveţi în îngrijire pacienţi minori care au fost diagnosticaţi cu o formă de cancer sau/şi care primesc în acest moment un tratament împotriva cancerului. Informaţiile pe care ni le veţi furniza au ca scop să ne ajute să înţelegem mai bine procesul de luare a deciziilor şi de a stimula dezbaterile pe tema autonomiei copiilor şi adolescenţilor cu privire la deciziile referitoare la starea lor de sănătate.

În cursul acestui interviu, vom încerca să cunoaştem starea de sănătate a pacientului dumneavoastră, calitatea şi conţinutul procesului de luare a deciziilor, de a avea acces la informaţii referitoare la capacitatea de decizie a pacientului minor. După interviu avem rugămintea să acceptaţi să completaţi un scurt chestionar, ce nu va depăşi o pagină. Vă asigurăm că răspunsurile dumneavoastră şi mai mult decât atât, toate informaţiile şi datele

furnizate vor fi tratate anonimizat şi în regim strict confidenţial. Vă mulţumim pentru că aţi acceptat să luaţi parte la acest studiu şi să ne răspundeţi la întrebări.

***Notă personală destinată persoanei ce va conduce interviul: în ceea ce priveşte întrebările marcate cu *, avem rugămintea de a le adapta în funcţie de răspunsurile oferite anterior. Spre exemplu, în ceea ce priveşte întrebările 2b şi c, dacă informaţiile au fost comunicate în acelaşi timp ambelor părţi, formulaţi întrebarea 2c în consecinţă; în cazul în care informaţiile au fost comunicate separat celor două părţi, vă rugăm să reformulaţi întrebarea.**

1. **Informaţii generale cu privire la pacient/caz:**
   a. De când cunoaşteţi pacientul şi familia sa?
   b. Când aţi văzut pacientul pentru prima dată şi când aţi pus diagnosticul?
   c. Ce vârstă are pacientul în prezent?
   d. Care este diagnosticul şi prognosticul?
   e. Cui aţi comunicat diagnosticul şi prognosticul? Dacă pacientul dumneavaostră nu era prezent, vă rugăm să ne spuneţi motivele pentru aceasta.
   f. Cum au reacţionat părinţii în momentul comunicării diagnosticului şi prognosticului?
   g. Dacă aţi discutat despre diagnostic cu pacientul minor, ne puteţi spune care a fost reacţia sa?
   h. Dacă aţi discutat despre prognosticul bolii cu pacientul minor, ne puteţi spune dacă i-aţi explicat şansele de vindecare?
   i. Dacă nu aţi discutat despre prognosticul bolii cu pacientul minor, ne puteţi spune care sunt motivele pentru aceasta?

j. Cine a iniţiat discuţia cu privire la diagnostic şi prognostic cu părinţii sau/şi pacientul minor? Cum v-aţi simţit în timpul acestei discuţii?

k. Puteţi să îmi descrieţi circumstanţele şi conţinutul acestei discuţii?

2. Opţiuni terapeutice, discuţii şi luarea unei decizii

a. Care au fost opţiunile terapeutice?

b. Cu cine aţi discutat aceste opţiuni terapeutice?*

c. Cum aţi descris opţiunile terapeutice pacientului minor/părinţilor?*

d. V-aţi simţit în largul dumneavoastră în timpul discuţiilor cu privire la opţiunile terapeutice? Am rugămintea să detaliaţi.

e. Există alternative de tratament în acest caz? Am rugămintea să precizaţi care sunt acestea.

f. Care au fost opţiunile rezonabile pentru un tratament clinic?

g. Aţi discutat despre avantajele şi inconvenientele acestei opţiuni de tratament? De ce da sau de ce nu?

h. După părerea dumneavoastră, în ce măsură a înţeles pacientul minor şi/sau părinţii acestuia opţiunile de tratament pe care le-aţi prezentat?*

i. Aceste discuţii au fost purtate cu pacientul minor şi/sau cu părinţii acestuia? Vă rog să precizaţi cine a fost prezent. Dacă pacientul minor a fost exclus de la aceste discuţii, vă rog să explicaţi din ce motive.

j. Ce opţiune de tratament a fost aleasă?

k. Cine a fost implicat în procesul de luare a acestei decizii?

l. Cum au fost luate deciziile cu privire la boala şi tratamentul pacientului minor? (Spre exemplu, care au fost punctele centrale ale discuţiilor, în ce manieră s-a ţinut cont de părerea persoanelor prezente şi cum s-a ajuns la un consens?)

m. În ce măsură a fost implicat pacientul minor în aceste discuţii? Am rugămintea de a preciza motivele pentru care copilul/adolescentul a fost/nu a fost implicat în luarea deciziilor.

n. În ce măsură aţi contribuit dumneavoastră la luarea deciziilor?

o. Experienţa şi părerea dumneavoastră au fost solicitate? Cât de comfortabil v-aţi simţit în legtură cu acesta?

p. Dumneavoastră aţi fost mulţumit/ă de rezultatul acestor discuţii?

q. Am rugămintea să precizaţi care sunt elementele ce au contribuit la un nivel de satisfacţie mai mare sau mai mic în legatură cu rezultatul discuţiilor.

r. În retrospectivă, aţi dori să schimbaţi ceva cu privire la aceste discuţii? Spre exemplu, există elemente care, după dumneavoastră, ar fi putut să fie benefice pentru discuţie/ ar fi putut-o ameliora?

3. **Implicarea copilului/adolescentului (sau excluderea din) în cadrul discuţiei:** Întrebările ce urmează se referă la capacitatea pacienţilor minori de a lua decizii cu privire la starea lor de sănătate şi la tratamentul medical, în special când suferă de o boală gravă. În prezent există o dezbatere în mediul medical şi public cu privire la implicarea copiilor şi/sau adolescenţilor în astfel de discuţii şi în ce masură aceştia au capacitatea şi posibilitatea de a participa la luarea deciziilor referitoare la starea lor de sănătate. Din aceste motive, ne interesează punctul dumneavoastră de vedere pe această temă.

a. În discuţia anterioară mi-aţi spus că la discuţiile legate de tratament pacientul minor a fost prezent (absent). Îmi puteţi explica motivele pentru aceasta?

b. De ce credeţi că a fost necesar sau potrivit îl implicaţi în (excludeţi din) cadrul acestor discuţii?

c. În ce condiţii aţi implica (exclude) pacientul minor din asftel de discuţii? Doriţi să precizaţi aceste condiţii mai detaliat, vă rog?

d. În general, când părinţii îşi implică (exclud) copiii din astfel de discuţii, care credeţi că sunt motivele pentru astfel de atitudini (aceste decizii)? De exemplu: ce criterii, valori menţioneaza ca stând la baza acestor decizii?

e. După părerea dumneavoastră pacientul minor ar trebui să aibă dreptul de a participa la luarea unor decizii ce privesc starea sa de sănătate? Vă rog să precizaţi motivele dumneavoastră (de ce sunteţi de acord sau de ce nu?)

f. După părerea dumneavoastră, care ar trebui să fie rolul părinţilor în cadrul procesului de luare a deciziilor?

g. După părerea dumneavoastră, pacienţii minori ar trebui să aibă dreptul, la modul general, de a lua decizii în ceea ce priveşte starea lor de sănătate? Vă rog să explicaţi mai detaliat răspunsul dumneavoastră:

- Răspunsul dumneavoastră ar fi la fel sau diferit în funcţie de:
    - Vârsta copilului: dacă are mai puţin sau mai mult de 8 ani (sau o altă limită a vârstei)?
    - Tipul şi gravitatea condiţiei medicale: (în funcţie de şansele de vindecare)
    - Comportamentul copilului
    - Religia sau convingerile dumneavoastră personale

**********MULŢUMESC**********

Dragă participant/ă: Vă mulţumim în avans pentru dorinţa de a completa cheastionarul următor:

C. Informaţiile demografice ale doctorilor:

i. Vârsta: _____ Anul naşterii: _____

j. Sex: ☐ Masculin ☐ Feminin

k. Naţionalitate: ☐ Română ☐ Alta – Ţara de origine: _____

l. Limbi vorbite: ☐ Româna ☐ Maghiara ☐ Germana ☐ Franceza

☐ Engleza ☐ Alta-precizaţi: _____

m. Starea civilă: ☐ Căsătorit/ă ☐ Divorţat/ă ☐ Văduv/ă

☐ Necăsătorit/ă ☐ Alta- Precizaţi _____

f. Specialitatea medicală: _____

g. Experienţa în oncologia pediatrică (în ani):

☐ 0 – 4 ani ☐ 5 – 8 ani ☐ 9 – 12 ani ☐ peste 12 ani

h. Religia: ☐ Ortodoxă ☐ Catolică ☐ Protestantă

☐ Alta- Precizaţi _____

**********MULŢUMESC**********

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

Interview Guide for Parents – Swiss study

English version

Dear participant: Thank you for agreeing to take part in this study and for sharing your information with us. You are being asked to take part in the research study of "**Attitudes and motives concerning end-of-life decisions: Competency and autonomy of children and adolescents in pediatric oncology**" because your child was diagnosed with cancer and/or is currently undergoing cancer treatment in a Swiss Pediatric Oncology Centre. We understand that this is a very sensitive issue and we would seek to be as considerate as possible. If you feel uncomfortable responding to any question, please communicate this to me.

During this interview session, we would like to learn about the condition of your child and the quality, content, and process of decision making regarding his/her illness. The information that we gain from you will help us understand decision making processes and contribute towards debates concerning a minors' decision making autonomy concerning health care. After the interview, we will request you to complete a very short 1 page survey. Please be assured that your responses and all the information/data you provide are completely confidential and anonymous.

1. Understanding the situation:

a. Let me start by asking you about your family. Could you tell me about them?

b. Please briefly describe to me the circumstances regarding your child's cancer diagnosis?

c. How did you first come to believe that your child might have cancer?

d. When was the diagnosis made? How old is your child now?

e. What was the prognosis the physician gave you?

***(Note to the Interviewer: Ask the following two questions only if the parent described the chances as poor.)**

1. When did you understand that the chances of cure for your child are rather low?

2. Could you please tell me more about how you see the chances of cure for your child?

f. Was your child present during the diagnosis discussion? Why or why not?

(*Note: If child present, ask question g, else skip to h.)

g. How did you feel about your child's presence?

h. Since the diagnosis was made, have you informed your child about the diagnosis?

i. How did you do so? Could you tell me the words you used? For example: what was the first sentence you used to start the discussion? Why did you choose the words you chose?

j. Did you discuss the prognosis with your child? Did you explain him/her the chances of cure?

k. Who initiated this discussion? How did you feel during the discussion?

2. Treatment options, discussions, and decision making

a) What were the treatment options?

b) Who were present when treatment options were discussed?

c) How were these treatment options described to you and/or your child by your physician?

d) Were these options clearly and carefully explained to you and/or your child by your physician?

e) If your child was not present during treatment discussions, have you discussed treatment or non-treatment options with your child?

(*Note: if answer is no, skip to question g.)

f. What did/do you think about this discussion with your child?

g. What treatment options were selected? And why?

h. How were decisions made about your child's illness and treatment choices?

i. Who was involved in this decision making process?

j. To what extend was the child involved? Please provide reasons for extent of involvement or non-involvement.

k. How comfortable were you with the manner and content of these discussions?

l. Were you satisfied with these discussions?

m. Please explain what factors contributed to greater or lower satisfaction.

a. Would you change anything retrospectively? e.g. Is there anything in your opinion that would have made these discussions more beneficial?

3. Inclusion or Exclusion of child in discussion: The following questions relate to the capacity of children to make decisions concerning their health and medical treatment, in particular, if they are severely ill. In society there is a debate about whether and how much children or minors should be involved and given the right to make decisions. We, therefore, would like to gather your opinion on this topic.

a) In your previous response, you stated that your child was present (absent) during treatment discussions. Could you explain the reasons for this?
b) Why do you think it was necessary/appropriate to include (exclude) your child from these treatment decisions?
c) Under what conditions would you absolutely include (exclude) your child in such discussions? Please explain these conditions.
d) Generally, when parents include (exclude) children from making such decisions, what kind of reasons, for example values and attitudes, are associated with it?
e) In your opinion, should children in general, be allowed to make health care related decisions? Please explain your perspective.
    - Would your response be the same or differ based on the following:
        - Age of the child: younger than 8 years or older than 8 years (or another age limit?)
        - Type and seriousness of the disease: terminal or non-terminal
        - Behaviour of the child
        - Your religion or personal beliefs

**********Thank You **********

Dear Participant: We thank you for taking the time to complete the following.

A. Demographic Information:

n. Age: Year of Birth: _____

o. Sex: ☐ male ☐ female

p. Nationality: ☐ Swiss ☐ Other – Country of origin: _____

q. Languages: ☐ German ☐ French ☐ Italian

☐ English ☐ Other (specify): _____

r. Marital Status: ☐ Married ☐ Divorced ☐ Widowed

☐ Never Married ☐ Other – Specify _____

s. Religion: ☐ Catholic ☐ Protestant

☐ Other (specify): _____

t. Number of children:

u. Education: ☐ No qualification ☐ Vocational ☐ High School

☐ College Degree ☐ University/Graduate Education

v. Living situation: ☐ Alone with children ☐ with spouse and children

☐ Other – specify:_____

**********Thank You **********

Curriculum Vitae

DOMNITA OANA BADARAU

Home Address: BASEL, CH-4056, Switzerland
Work Address: Bernoullistrasse 28, BASEL, CH-4058, Switzerland
E-mail: domnita.badarau@case.edu; do.badarau@gmail.com

PROFILE

Professional specialized in research ethics and European and US regulatory frameworks for research with human participants, with expertise in compliance for (cross-cultural) clinical studies, with over 7 years of interdisciplinary research at the intersection of health, law and research, project and grant management, trained in Good Clinical Practice and research processes, with working experience in ethics approval, clinical study documentation, coordinating communication with diverse teams and clinicians, and over 2 years' practice in financial and legal compliance, risk assessment and loan applications, control and administration.

PROFESSIONAL EXPERIENCE

A. RESEARCH

Clinical Study Coordinator / Postdoctoral Research Associate (2017 – present), Aston University, School of Life and Health Sciences and Aston Brain Centre; Birmingham, United Kingdom

- Coordinate scientific and clinical study activities for PROBIt, a multidisciplinary study of children's brain development aimed at improving clinical practice via evidence-based research in the area of long-term neurobehavioral outcomes prediction in children with brain insults
- Project management for all study phases (from planning and design, governance, recruitment strategies and participant enrollment, data collection, maintenance, anonymization, site files and secure storage, reporting to dissemination of results)
- Lead submissions to Health Research Authority and support ethics requirements for all work packages: preparing and submitting documentation, amendments, new applications, annual reporting and adverse event reporting
- In charge of critical communication and collaboration with academic, clinical and research organizations for study related activities and science communication
- Manage the research staff, supervise and co-supervise students
- Plan study activities and delivery, and support the Chief Investigator in all study administrative and research matters, as well as the PIs at each collaborating site
- Liaise with and act as a key contact for all study stakeholders, including the funder (European Research Council), Scientific Steering Committee, collaborators, clinicians, research team and participating families
- Maintain records of study budget, including invoicing and ensure accuracy; study audit documents and supervise purchase orders/expenditures
- Carry systematic literature searches and review scientific databases, summarize findings, register reviews and prepare reports and submit outputs of the study
- Support impact activities, including collating information and disseminate to academic and community stakeholders

Honorary research staff (2017 – present), Department of Neurophysiology, Birmingham Women's and Children's NHS Foundation Trust; Birmingham, United Kingdom

- Support and coordinate research activities (scanning sessions and cognitive assessments) and study recruitment/enrolment
- Act as key contact for the PROBIt study and coordinate medical data collection and transfer

Clinical Trials Trainee / Postdoctoral Research Collaborator (2017 – present), Department of Medicine, Swiss Tropical and Public Health Institute; Basel, Switzerland

- Conduct research on legal and regulatory frameworks impacting clinical trials in resource constrained settings
- Support current projects on ethico-legal issues arising from dealing with high uncertainty in recruitment and informed consent procedures, with a focus on pediatric studies
- Data mining of scientific databases and past STPH project repositories
- Develop a database (knowledge pool) on research ethics review processes, from planning to approval and implementation, including customary practices regarding consent; Project focused on Sub-Saharan Africa and Central Asia
- Collaborate on study recruitment and participant documentation design
- Conduct systematic literature searches, register reviews and submit outputs for dissemination (manuscripts and scientific conferences)

Research Assistant (2011 – 2016), University of Basel, Institute for Biomedical Ethics (IBMB); Basel, Switzerland

PROJECTS

(1) **Communication and patient involvement in pediatric oncology care in Romania** (a PhD project conducted in Romania and Switzerland; funded by the the Botnar Stiftung, Switzerland)

The study explored parents' and physicians' experiences in communicating with minor patients about their cancer diagnosis, identified barriers to diagnosis disclosure and examined the different ways children and adolescents are supported or hindered to participate in their care. Qualitative interviews were employed for data collection.

(2) **Attitudes and motives concerning end-of-life decisions: Competency and autonomy of children and adolescents in pediatric oncology** (prospective 3 years project funded by the Swiss National Science Foundation (SNSF), Switzerland)

The study investigated parents' and oncologists' attitudes and practices on minor patients' involvement in care. Children's and adolescents' views on their inclusion during treatment for cancer were also examined. Data were collected in the German, French and Italian-speaking parts of Switzerland through quantitative surveys and qualitative interviews.

RESPONSIBILITIES

1. Conduct research in the area of pediatric oncology and ethics

- Project management for all study phases (from planning, participant enrollment, data collection, maintenance, de-anonymization, secure storage, analysis to dissemination)

- Managed ethics approval – preparation of study documentation: writing informed consent documents, participant information sheets, and monitoring of all documents
- Performed study monitoring and reporting, including to funding agencies and ethics committees
- Designed and implemented a study in university hospitals in Romania (PhD project)
- Co-investigator for a Swiss-wide study in 8 cantons (German, French and Italian); collaborated in study design and implementation
- Conducted 28 one-on-one in-depth interviews with research subjects
- Advised on building questionnaires, interview guides and participant information sheets and reviewed them
- Developed and expanded collaborations with physicians/clinics; ensured communication with 11 clinical institutions, collaborating researchers and diverse organizations (funding agencies, compliance institutions)
- Drafted/edited and translated reports, research and other documents from English to French
- Disseminated research findings in scientific publications – 7 original articles – and presented findings at international conferences, professional meetings

2. Advise on and support for diverse research studies

- Advised on study design/protocols, ethics issues for projects in public health, fertility preservation, pediatric palliative care and prisoner health issues
- Reviewed grant proposals and PhD projects
- Performed peer-review of scientific manuscripts, conference abstracts, commentaries authored by colleagues

COMMUNICATION AND RESEARCH ETHICS TRAINING

1. Manage teaching activities

- Developed and taught courses and one-day trainings in compliance, research ethics and clinical ethics (Medical Faculty, University of Basel)
- Evaluated and commented on students' research presentations on public health
- Tutored students in the Medical and Science Faculties on research legal regulations for research with human subjects and ethical principles

2. Coordinate courses and public lectures on research compliance and ethics

- 1 elective semester course, 2 lecture visits and workshops for Prof. Sana Loue (US) and Prof. Ezekiel Emanuel (former director of NIH Department of Bioethics, US)
- Co-organization of the 27th European Conference on Philosophy of Medicine and Healthcare (ESPMH), (August 2013)

NIH Fogarty International Fellow (August 2010 - January 2012)

Case Western Reserve University, School of Medicine; Cleveland, United States

1. Research ethics

- Participated in and observed daily activities of regulatory bodies (Institutional Review Boards and ethics committees' meetings); drafted post-meeting reports
- Revised, improved quality and reduced readability of informed consent documents and

participant recruitment tools

- Performed critical methodological, ethical and legal analysis of clinical trials (focus on research in African countries)

2. Clinical ethics expertise

- Performed participatory observational work and shadowed medical round in various medical units within Case Medical Center network (100 hours) to train and gain expertise in clinical ethics
- Participated in clinical case consultation as an observer and performed patient case analysis / drafted final reports
- Compiled notes from medical rounds and ethics committees meetings

B. Legal and business sector

Legal, Control and Administrative Support Officer for small and medium enterprises (December 2008 – July 2010)

Raiffeisen Bank S.A., member of Raiffeisen Zentralbank Oesterreich (RZB); Romania

- Coordinated communication and carried out credit approvals by preparing and ensuring documentation compliance with Risk, Control and Admin Departments' regulations
- Managed the department's client portfolio by supporting and assisting the Relationship Manager in loan application processes
- Offered legal advice and law interpretation to internal departments

Junior Relationship Manager (March 2008 – December 2008)

Raiffeisen Bank S.A., member of Raiffeisen Zentralbank Oesterreich (RZB); Romania

- Trainee in banking operations, sales, financing and legal-financial analysis of documents for submitting loans applications to the Risk Department

Education

2012 – 2016	**Doctoral studies in medical sciences (PhD)** Institute For Biomedical Ethics, University of Basel, Basel, Switzerland Supervisors: Prof. Bernice S Elger, MD; Tenzin Wangmo, PhD; Prof. Thomas Kühne, MD; Prof. Sana Loue, PhD
2010 – 2011	**MA** in Bioethics Case Western Reserve University, School of Medicine, Department of Bioethics, United States
2008 – 2009	**MIB**, International Commerce Faculty of Economics and Business Administration, Al. I. Cuza University, Romania
2004 – 2008	**Bachelor of Economics and Business Administration**, specialization in International Economic Relations Faculty of Economics and Business Administration, Al. I. Cuza University, Romania
2004 – 2009	**Bachelor of Law**

	Faculty of Law, Al. I. Cuza University, Romania
September 2007 – February 2008	**Erasmus** student in the Lifelong Learning Programme Faculty of Economics IQS, Ramon Llull University, Barcelona, Spain

Relevant trainings, workshops and seminars

November 2015	**Zürich Intensive Ethics Course,** University Hospitals Zürich, University of Zürich, Harvard Medical School, Zürich, Switzerland
April 2015	**Research Methods in Bioethics – Data analysis**, University of Basel, Switzerland
October 2014	**ESO/SEMM Master in Ethical Counseling in Oncology**, European School of Molecular Medicine, Milan, Italy
June 2014	**Qualitative methods course for bioethicists**. **Summer School**– University of Zürich, Zürich, Switzerland
August 2013	**Summer School Lugano** - ***Advanced Methods in the Social Sciences***, University of Lugano, Lugano, Switzerland
April 2013	**Advanced Research Methods in Bioethics** – specialized topics, University of Basel, Basel, Switzerland
October 2012	**Health Systems,** Swiss Tropical Public Health Institute, Basel, Switzerland

Consulting

2015	Consultant for evaluation of participant/patient research documents with Center for Information and Study on Clinical Research Participation (CISCRP), US

Professional Affiliations, Special Appointments, Grants

2017 – onward	Member of the Editorial Board of the Journal of Immigrant and Minority Health, Springer Science+Business Media New York
2012 – 2016	Hemmi Stiftung, Basel, Switzerland
2015 – 2016	Botnar Stiftung, University of Basel, Switzerland
2015	ANTELOPE career-program for young female researchers, University of Basel, Switzerland
2010 – 2012	Fogarty Fellowship, International Research Ethics Training Program, Fogarty International Center, National Institutes of Health, US
2007 – 2008	Erasmus scholarship, Erasmus LLP, Ramon Llull, Barcelona, Spain, awarded by the European Union

Publications

A. Peer-reviewed articles

1) **Domnita O Badarau**, Eva De Clercq, Bernice S Elger (2019). Continuous deep sedation and euthanasia in pediatrics. Does one really exclude the other for terminally ill patients? The Journal of Medicine and Philosophy

2) **Domnita O Badarau**, Katharina M Ruhe, Thomas Kühne, Eva De Clercq, Anca Colita, Bernice S Elger, Tenzin Wangmo (2017). Decision making in pediatric oncology: Views of

parents and physicians in two European countries. AJOB Empirical Bioethics

3) **Domnita O Badarau**, Eva De Clercq, Tenzin Wangmo, Monica Dragomir, Ingrid Miron, Thomas Kühne, Bernice S Elger (2016). Cancer care in Romania: challenges and pitfalls of children's multifaceted involvement. Journal of Medical Ethics

4) Katharina M Ruhe, Tenzin Wangmo, Eva De Clercq, **Domnita O Badarau**, Marc Ansari, Thomas Kühne, Felix Niggli, Bernice S Elger; Swiss Pediatric Oncology Group (2016). Putting patient participation into practice in pediatrics – Results from a qualitative study in pediatric oncology. European Journal of Pediatrics

5) Tenzin Wangmo, Katharina M Ruhe, **Domnita O Badarau**, Thomas Kühne, Felix Niggli, Bernice S Elger; Swiss Pediatric Oncology Group (2016). Parents' and patients' experiences with pediatric oncology care in Switzerland – Satisfaction and some hurdles. Swiss Medical Weekly

6) Katharina M Ruhe, **Domnita O Badarau**, Pierluigi Brazzola, Heinz Hengartner, Bernice S Elger, Tenzin Wangmo; Swiss Pediatric Oncology Group (2016). Participation in pediatric oncology: views of child and adolescent patients. Psycho-Oncology

7) **Domnita O Badarau**, Tenzin Wangmo, Katharina M Ruhe, Ingrid Miron, Anca Colita, Monica Dragomir, Jan Schildmann, Bernice S Elger (2015). Parents' challenges and physicians' tasks in disclosing cancer to children. A qualitative interview study and reflections on professional duties in pediatric oncology. Pediatric Blood & Cancer

8) Katharina M Ruhe, Tenzin Wangmo, **Domnita O Badarau**, Bernice S Elger, Felix Niggli (2015). Decision-making capacity of children and adolescents - suggestions for advancing the concept's implementation in pediatric healthcare. European Journal of Pediatrics

9) Eva De Clercq, **Domnita O Badarau**, Katharina M Ruhe, Tenzin Wangmo (2014). Body matters: rethinking the ethical acceptability of non-beneficial clinical research with children. Medicine, Health Care and Philosophy

10) Katharina M Ruhe, **Domnita O Badarau,** Bernice S Elger, Tenzin Wangmo (2014). End-of-Life decision making in pediatrics: Literature review on children's and adolescents' participation. AJOB Empirical Bioethics

B. Abstract(s)

1) **Domnita O Badarau**, Anca Colita, Monica Dragomir, Bernice S Elger, Thomas Kühne, Ingrid Miron, Felix Niggli, Katharina M Ruhe, Tenzin Wangmo (2014). Decision-making for children following a cancer diagnosis – Preliminary findings from a qualitative study with parents and oncologists. Pediatric Blood & Cancer (Supplement S2)

C. Book chapter(s)

Domnita O Badarau, Beatrice G Ioan (2013). Forensic Epidemiology in the Romanian Legal System. In S. Loue (Ed.), Forensic Epidemiology in the Global Context (pp. 79-98): Springer New York

D. Commentary and Encyclopedias

1) **Domnita O Badarau**, Rebecca L Nast, David M Shaw (2014). The vulnerability of the individual benefit argument. American Journal of Bioethics

2) **Domnita O Badarau** (2013). Declaration of Helsinki. In S. Loue (Ed.), Mental Health

Practitioner's Guide to HIV/AIDS. Springer New York

3) **Domnita O Badarau** (2013). Disclosure Laws. In S. Loue (Ed.), Mental Health Practitioner's Guide to HIV/AIDS. Springer New York
4) **Domnita O Badarau** (2013). Economic Impact. In S. Loue (Ed.), Mental Health Practitioner's Guide to HIV/AIDS. Springer New York
5) **Domnita O Badarau** (2013). Human Trafficking. In S. Loue (Ed.), Mental Health Practitioner's Guide to HIV/AIDS. Springer New York
6) **Domnita O Badarau** (2013). Informed Consent. In S. Loue (Ed.), Mental Health Practitioner's Guide to HIV/AIDS. Springer New York
7) **Domnita O Badarau** (2013). World Trade Organization. In S. Loue (Ed.), Mental Health Practitioner's Guide to HIV/AIDS. Springer New York
8) **Domnita O Badarau** (2012). Asylum. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA
9) **Domnita O Badarau** (2012). Brain Drain. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA
10) **Domnita O Badarau** (2012). Ghetto. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA
11) **Domnita O Badarau** (2012). Guest Worker. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA
12) **Domnita O Badarau** (2012). Hague Convention on Child Abduction. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA
13) **Domnita O Badarau** (2012). Labor. Encyclopedia of Immigrant Health, Springer Science+Business Media, LLC, USA

Presentations

A. Invited oral presentations

1) **Domnita O Badarau** (2016). What to consider when breaking bad news? Ethical challenges in communication in pediatric oncology, 12th International Bernd-Spiessl-Symposium, University Hospitals Basel, Switzerland
2) **Domnita O Badarau** (2015). Between finding their voice and giving children a voice. Parental struggles in pediatric oncology. Case Western Reserve University, School of Medicine, Department of Bioethics, Cleveland, US

B. Oral presentations

3) Katharina M Ruhe, Eva De Clercq, **Domnita O Badarau**, Bernice S Elger, Tenzin Wangmo (2015). Competency and autonomy of children and adolescents in pediatric oncology. SPOG Scientific Meeting, Lugano, Switzerland
4) Katharina M Ruhe, **Domnita O Badarau** (2014). Competency and autonomy of children and adolescents in pediatric oncology. A study update. SPOG Scientific Meeting, Lugano, Switzerland
5) **Domnita O Badarau**, Anca Colita, Katharina M Ruhe, Tenzin Wangmo, Bernice S Elger (2013). Competency and autonomy of children and adolescents in pediatric oncology decision-making. The 4th EBMT Training Course for Paediatricians and Paediatric Nurses on HSCT in Children and Adolescents: Interactive Educational EBMT PDs Course; ESH.EBMT, Bucharest, Romania

6) Katharina M Ruhe, **Domnita O Badarau**, Tenzin Wangmo, Bernice S Elger (2013). Behandlungsentscheidungen in der pädiatrischen Onkologie und die Einbeziehung von Kindern und Jugendlichen – Vorläufige Ergebnisse einer qualitativen Studie mit Patienten und Eltern. Fachtagung Psychoonkologie, St. Gallen, Switzerland

7) Katharina M Ruhe, **Domnita O Badarau**, Bernice S Elger, Tenzin Wangmo (2013). The inclusion of children in decision-making at the end-of-life: A review of the literature. European Association of Centers of Medical Ethics Annual Conference, Bochum, Germany

8) **Domnita O Badarau**, Tenzin Wangmo, Katharina M Ruhe, Anca Colita, Bernice S Elger (2013). Is it ethically justifiable to involve children in non-therapeutic research? Bioethics, Medical Ethics & Health Law. Towards the 21st century, Naples, Italy

C. Poster presentations

9) **Domnita O Badarau**, Anca Colita, Monica Dragomir, Bernice S Elger, Thomas Kühne, Ingrid Miron, Felix Niggli, Katharina M Ruhe, Tenzin Wangmo (2014). Decision-making for children following a cancer diagnosis – Preliminary findings from a qualitative study with parents and oncologists. SIOP, Toronto, Canada

10) Katharina M Ruhe, **Domnita O Badarau**, Bernice S Elger, Felix Niggli, Thomas Kühne, Tenzin Wangmo (2013). Children's and Adolescents' Experiences in Pediatric Oncology Treatment Decision Making. 15th World Congress of Psycho-Oncology and Psychosocial Academy, Rotterdam, Netherlands

11) **Domnita O Badarau**, Katharina M Ruhe, Tenzin Wangmo, Bernice S Elger (2013). Blurred lines in pediatric oncology. National Research Conference on Palliative Care, Swiss Academies of Arts and Sciences, Bern, Switzerland

12) Katharina M Ruhe, Tenzin Wangmo, **Domnita O Badarau**, Bernice S Elger (2013). Physicians' attitudes towards patient participation in decision making in the pediatric oncology setting and their assessment of competency-Preliminary findings from a qualitative study. European Cancer Conference, Amsterdam, Netherlands

13) **Domnita O Badarau**, Bernice S Elger, Thomas Kühne, Tenzin Wangmo (2012). Acute and chronic pain management: An issue of concern in pediatric oncology. 80th Annual Meeting, Swiss Society of General Internal Medicine, Basel, Switzerland

14) Tenzin Wangmo, **Domnita O Badarau**, Bernice Elger (2012). Children's autonomy in making medical end-of-life decisions. Thinking Ahead – 11th World Congress of Bioethics, International Association of Bioethics. Rotterdam, The Netherlands

Teaching

39429-01: 1h presentation on "Legal and ethical aspects of capacity in minor patients" as part of the Contemporary Debates in Bioethics lecture series on Reproductive ethics (Spring 2013)

Course coordinator of the Contemporary Debates in Bioethics lecture series, IBMB, University of Basel (Spring 2012)

Group seminar: Co-tutor for the Health, Personal Responsibility, Social Exclusion/Lebenszyklen, Medical School, University of Basel (Spring 2013)

Small-group seminar: co-tutor in the Psychology-Ethics-Law (PoT Problem oriented Tutoring)/Psyche-Ethik-Recht (PER I), Medical School, University of Basel (Spring 2012)

30209-01: commentary on research presentation in the course Ethics in Biomedical and

Public Health Research, IBMB, University of Basel (Spring 2012)

10423-01: Tutor for the Introduction to ethics/Grundlagen der Ethik, Bio- and Pharmacology Faculty, University of Basel (Spring 2012)

Small-group seminar: organization and tutor in the Body, Subject, Environment/Körper, Subjekt, Umwelt block course, Medical School, University of Basel (Fall 2011)

28724-01: 2h presentation as part of Biomedical Ethics seminar, IBMB, University of Basel (Fall 2011)

Languages

English - Proficient
German - Intermediate
Spanish - Proficient
French - Intermediate
Romanian - Native
Italian - Elementary

RESEARCH INTERESTS

Research ethics and pediatric clinical trials, MRI pediatric research, pediatric ethics, parental experiences, child and adolescent participation in care, communication in pediatric oncology, decision making, (international) research ethics, cultural sensitivity

Interests

- Reviewer for international scientific journals (Journal of Immigrant and Minority Health, Journal of Pediatric Nursing, Journal of Empirical Research on Human Research Ethics).
- French classes and visiting international development projects in in Burkina Faso, West Africa, in 2016-2017.
- Volunteer with local Basel organization (OESA) for the Kid's Playgroup at a center for asylum seekers, Switzerland; working with children from Congo, Syria, Afghanistan, Iraq.
- Volunteer with regional group (NEOhcn) to serve disadvantaged communities in a city with diverse racial make-up, Cleveland, US.

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