Paediatric **E**nd-of-**LI**fe **CA**re **N**eeds in Switzerland (**PELICAN**): Current end-of-life care practices and the perspectives of bereaved parents

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Table of Contents

List of abbreviations

BPNA	Bereaved Parent Needs Assessment
C	Cardiology
CAD	Central Access Device
CASC	Comprehensive Assessment of Satisfaction with Care
CCC	Complex Chronic Condition
CPR	Cardiopulmonary Resuscitation
DNR	Do not Resuscitate
EAPC	European Association for Palliative Care
ECMO	Extracorporeal Membrane Oxygenation
EFA	Exploratory Factor Analysis
EOL	End-of-Life
GEE	Generalized Estimating Equations
HCT	Health Care Team
ICD	International Statistical Classification of Diseases and Related Health Problems
I-CVI	Item-Content Validity Index
ICU	Intensive Care Unit
IPPC	Initiative for Pediatric Palliative Care
IQR	Interquartile Range
ISAT	Institutional Self-Assessment Tool
ISPOR	International Society of Pharmacoeconomics and Outcome Research
KMO	Kaiser-Meyer-Olkin
MSA	Measure of Sampling Adequacy
N	Neurology
NA	Not Applicable
Neo	Neonatology
NPQ	Needs of Parents Questionnaire
NICU	Neonatal Intensive Care Unit
O	Oncology
PaPEQu	Parental PELICAN Questionnaire
PC	Palliative Care
PICU	Paediatric Intensive Care Unit
PPC	Paediatric Palliative Care
QOL	Quality of Life
S-CVI	Scale-Content Validity Index
WHO	World Health Organization

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"The pelican (*Henet* in **Egyptian**) was associated in **Ancient Egypt** with death and the **afterlife**. It was depicted in art on the walls of tombs, and figured in funerary texts, as a protective symbol against snakes. References in non-royal funerary papyri show that the pelican was believed to possess the ability to prophesy safe **passage in the underworld** for someone who had died."

Wikipedia the free encyclopedia. *Pelican*. 01.04.2016, [accessed 15.08.2012].

Karin Zimmermann, April 2016

Summary

Despite continued advancements in medical care and improved survival or life expectancy, childhood deaths due to complex chronic conditions (CCC) or prematurity are inevitable. Deaths during the first year of life constitute approximately 50% of disease-related deaths, the causes of which include perinatal complications, prematurity, or congenital anomalies. Beyond the age of one year, the three most common life-limiting CCCs are neurological/neuromuscular and cardiovascular conditions (including genetic disorders), and malignancies. The majority of disease- and prematurity-related deaths occur in hospitals and for children dying at home, hospital use in their terminal stage is high. Symptom burden and reliance on medical technology has been reported to be considerable. Circumstances and characteristics of deaths, however, are known to vary by age and medical conditions.

When facing the death of their child, parents experience an unimaginably painful life event and severe crisis that affects the whole family for life. In this highly stressful time parents are confronted with uncertainty and are required to make difficult decisions, e.g. withdrawal of life-sustaining interventions. Their need for compassionate professional support is high. Paediatric palliative (PPC) and end-of-life (EOL) care emerged as a medical subspecialty aimed at meeting the specific needs of seriously ill children and their families. Meeting these needs requires a comprehensive and integrative approach from a compassionate and skilled multidisciplinary team. Parental needs have been studied in the past and an overview of themes/domains most important to parents can be summarised as: *sincere relationships and emotional, spiritual and cultural support*; *genuine communication*; *alleviation of suffering*; *continuity, coordination and accessibility of care*; and *bereavement support*. Deficiencies in meeting parental needs were identified across all themes, e.g. insufficient communication, lack of respect, and lack of emotional support.

Most of the evidence related to the parental perspective of their child's EOL care originates from qualitative research. To assess and explore the parental perspective effectively, an approach combining quantitative and qualitative information is likely to provide the most comprehensive view of experiences and unmet needs. A few self-administered questionnaires designed to assess the experiences and needs of parents of a dying child exist. None of the instruments available were applicable to the heterogeneous field of paediatric EOL care, including children of different age groups, with different underlying illnesses and in different clinical settings.

The development of PPC in Switzerland lags behind when compared with other developed countries, such as the United Kingdom or the USA. Need for action has been recognised by the Federal Office of Public Health and the call for a thorough assessment of current practices, needs, available resources and existing services as requirement to formulate and plan further measures to promote PPC in Switzerland has been issued.

This dissertation is embedded in the **PELICAN** study (**P**aediatric **E**nd-of-**LI**fe **CA**re **N**eeds in Switzerland, 2012 – 2015). The overarching aims of the nationwide PELICAN study were to provide comprehensive information and to understand the current practice of EOL care (i.e. in this study, the last 4 weeks of life prior to death) in paediatric settings in Switzerland (hospital and community care) and to explore and describe parental perspectives and the perspectives of the healthcare professionals involved. Based on the results, recommendations for best-practice during this highly vulnerable and critical phase of life not only for the affected child but also for the family will be formulated.

The dissertation is organized in **9 chapters**:

Chapter 1 introduces in the field of paediatric EOL care and reviews relevant topics. The epidemiology of childhood illness- and prematurity-related death is described together with characteristics and circumstances of death. PPC and EOL are being defined and the he provision of paediatric palliative and EOL care are introduced. Existing evidence about the parental perspective of their child's EOL care is summarised, and a potential framework for the quantitative assessment of parental experiences and needs is described. The emerging research gaps, and the rational and aims of this dissertation are described in **chapter 2** together with the context in which the PELICAN study took place.

Chapter 3 comprises the PELICAN study protocol. It provides an introduction, rational and specific aims of the entire research project including three main studies and two subs-studies

combining quantitative and qualitative methodology. Methods for each study arm are detailed. This dissertation comprises PELICAN I and the quantitative part of PELICAN II, including the development and testing of the Parental PELICAN Questionnaire (PaPEQu).

Chapter 4 reports on patterns of care at EOL in neonates and children with complex chronic conditions. Data from 149 paediatric patients who died in the years 2011 or 2012 due to a cardiac, neurological or oncological condition, or during the neonatal period were collected in 13 hospitals, two long-term institutions and 10 community-based healthcare service providers throughout Switzerland. Sixty-two percent of the patients died in intensive care units, 84% of them following the withdrawal of life-sustaining treatment. Reliance on invasive medical interventions was prevalent, and the use of medication was high, with a median count of 12 different drugs during the last week of life. Patients experienced an average number of 6.42 symptoms. The prevalence of various types of symptoms differed significantly among the four diagnostic groups. Only half of the patients who spent days at home received community-based healthcare. The study provides a comprehensive overview of current EOL care practices in a real-life setting of different healthcare providers and the findings provide a knowledge base for paediatric palliative care teams.

Chapter 5 describes the development and initial validation of the Parental PELICAN Questionnaire (PaPEQu) – an instrument to assess parental experiences and needs during their child's EOL care. The PaPEQu was developed in four phases between August 2012–March 2014: phase 1: item generation; phase 2: validity testing; phase 3: translation; phase 4: pilot testing. Psychometric properties were assessed after applying the PaPEQu in a sample of 224 bereaved parents in April 2014. Validity testing covered the evidence based on tests of content, internal structure and relations to other variables. The PaPEQu consists of approximately 90 items in four slightly different versions accounting for particularities of the four diagnostic groups. The questionnaire's items were structured according to six quality domains described in the literature. Evidence of initial validity and reliability could be demonstrated with the involvement of healthcare professionals and bereaved parents. The PaPEQu holds promise as a measure to assess parental experiences and needs and is applicable to a broad range of paediatric specialties and settings.

Chapter 6 reports on the results of the survey with the newly developed PaPEQu. Responses regarding parental experiences and perceived satisfaction are described. Differences between the four diagnostic groups are analysed using a generalized estimation equation to account for the dyadic data structure. Of 307 eligible families, 267 could be contacted and 135 (51 %) consented to participate in this questionnaire survey. Our findings show positive parental experiences of their child's EOL care and high perceived satisfaction with the care their child received. Parents of a child with cancer rated their experiences highest in most of the six quality domains and reported the highest satisfaction with care. The lowest scores were mainly reported by parents from the neurology group, with the exception of the shared decision making domain, where parents of neonates reported significantly less positive experiences. Although positive in general, our study results suggest some areas for improvement. The integration of specialised paediatric palliative care has the potential to minimise lost opportunities to support and assist parents.

In **chapter 7** finally, the results of all studies are being synthesized and discussed within the state of science. Strengths and weaknesses of methods are examined and implications on the level of policy, practice and research are derived. This dissertation concludes with the recognition that urgently needed data to advance the development of PPC in Switzerland is now available. It also recognises however, that we are challenged to demonstrate the effectiveness of PPC services in order to grow out of the infancy of this medical subspecialty.

Chapter 1
Introduction and Background

1.1. Setting the stage

Children living with life-limiting complex chronic conditions (CCC) and prematurely born babies are part of our health care system. And despite dramatic improvements in medical care over the last decades, death is not preventable for some children[1]. Death is part of life, but when it comes during childhood it is definitely out of season. Each child lost is a loss of human potential and living through the experience of losing a child might be considered as one of the most harrowing event in one's life. Dying and losing a child is accompanied by physical and emotional suffering and leaves a family wounded for life. All possible efforts have to be made to alleviate suffering of each dying child and to relieve the negative effects on families affected. Especially when death comes with some forewarning - as is mostly the case in children living with a life-limiting CCC and to some degree also in premature babies – nurses, physicians, social workers, psychologists and other healthcare professionals play an important role in the life of the dying child and his or her family [1]. At the same time, caring for a dying child and supporting his/her family is one of the most challenging situations for a health care team (HCT) [2]. High levels of expertise and knowledge of disease specific aspects of paediatric end-of-life (EOL) care are required. Equally important, professionals involved in paediatric EOL care need to be aware of the child's and his/her family's needs and should understand the perspectives of parents facing the death of their child in order to being able to provide best possible care. Current paediatric EOL care practices in Switzerland and the perspectives of bereaved parents are the main topics that will be addressed in this dissertation and the results of two observational studies including the development and testing of an instrument to quantitatively assess parental experiences and needs during their child's EOL care will be presented and discussed.

1.1.1. Paediatric Palliative Care (PPC)

The need and vision to provide compassionate professional care has led to the emergence of the medical subspecialty of palliative care (PC), which found its origin in the hospice movement for adults in the UK of the sixties [3]. Palliative is derived from the Latin term palliare, "to cloak" [4]. Terms like PC, comfort care, supportive care, EOL care, and terminal care are often used interchangeably and creating difficulties in differentiating the concepts of palliative care with other care models, such as the chronic care model or concepts, such as comfort care and supportive care in the oncology setting [5]. It has therefore been argued that in a definition for PC, a relation to death and dying should be recognisable [5]. The World Health Organization's (WHO) definition for PC could therefore be criticised in that regard as this relation is not implicit. Nevertheless, the WHO's definition is the one most commonly applied and, importantly, a separate definition for paediatric palliative care (PPC) is provided.

According to the World Health Organization (WHO) "palliative care is an approach that improves the quality of life of patients and their families facing the problem associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual" [6]. The definition of PC for children specifies as follows:

- "Palliative care for children is the active total care of the child's body, mind and spirit, and also involves giving support to the family.
- It begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease.
- Health providers must evaluate and alleviate a child's physical, psychological, and social distress.
- Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources; it can be successfully implemented even if resources are limited.

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[1] The term child is used as an umbrella term for newborns, infants, children and adolescents.

- It can be provided in tertiary care facilities, in community health centers and even in children's homes." [6].

1.1.2. Paediatric end-of-life care

More specifically and as part of PC, the term EOL care refers to care when death is imminent [4]. The timeframe of EOL care is not well defined and, depending on the source, is described as a period of one to two years, or some weeks or days [7, 8]. A definition for paediatric EOL care is provided by together for Short lives, the leading UK charity with the mission to "secure the best quality of life and best end of life care for children who will have short lives" [9]:

"End-of-life care is care that helps all those with advanced, progressive, incurable illness to live as well as possible until they die. It focuses on preparing for an anticipated death and managing the end stage of a terminal medical condition. This includes care during and around the time of death, and immediately afterwards. It enables the supportive and palliative care needs of both child/young person and the family to be identified and met throughout the last phase of life and into bereavement. It includes management of pain and other symptoms and provision of psychological, social, spiritual and practical support and support for the family into bereavement." [10].

1.2. Epidemiology

Fortunately, illness- and prematurity-related childhood deaths are rare events. In Switzerland, a total of 424 children (0 – 14 years) died in 2013. About 15% of those deaths are related to accidents, sudden infant death syndrome or unknown causes, leaving 361 deaths due to illness or prematurity, including incidents during or shortly after birth [11]. Deaths due to perinatal conditions, including prematurity, constitutes about 50% of all deaths during the first year of life [12]. Approximately three quarters of these infants die within 24 hours after birth [11]. Beyond the first year of life, life-limiting CCCs such as neurologic illnesses, including chromosomal anomalies and metabolic/endocrine conditions, together with neoplasms and cardiac/circulatory conditions, including congenital malformations, are mostly responsible for illness-related childhood deaths [11, 12]. Diagnostic study results from the United States (US)/Canada and the United Kingdom showed that the predominant conditions of children who received PPC services in 2008 or died in 2009/2010, respectively, were congenital/chromosomal (41%, 32%), neuromuscular (39%)/encephalopathy (22%), and cancer (20%, 18%) [13, 14]. A recent study from the United Kingdom estimating the burden of life-limiting CCCs in children (0 – 19 years) also found that their prevalence increased over 10 years from 25 to 32 per 10 000 population.

1.3. Characteristics and circumstances of childhood death

1.3.1. Place of death

Stemming mostly from studies undertaken in adults, there is a commonly accepted view that home is the preferred place of death for most patients [15]. Place of death in children with life-limiting CCCs has been studied in the past and one conclusion from a nationwide US study was that children increasingly die at home, but that, overall, 82% still died in the hospital [16]. Depending on age, the percentage of deaths at home increased over a 15-year period between 1989 and 2003 for infants (4.9% vs. 7.3), for 1– to 9-year-olds (17.9% vs. 30.7%), and for 10- to 19-year-olds (18.4% vs- 32.2%). This shift was mostly attributed to advances in medical home-based technology and attitudes towards PPC. The percentages of home deaths also varied among different types of CCCs. Children with malignancies were most likely to die at home and infants least likely [16]. For Europe, data from a population-based study conducted in Belgium, the Netherlands, Italy, Norway, England and Wales showed home death rates of between 20% and 29% in 1037 children deceased due to a life-limiting CCC and aged 1 to 17 years. Again, home death was more common in children with cancer and, independently, also more likely in children aged 10 years and above [17].

While it has been recognized that children often want to be at home and families want to keep them at home [17, 18], in reality, most children die in hospital and many in intensive care units (ICU) [19, 20]. ICUs in paediatrics are differentiated into neonatal ICUs (NICU) and paediatric ICUs (PICU). NICUs are specialised units caring for premature and ill newborns requiring intensive medical treatment. Critically ill newborns needing surgical interventions are usually transferred to a PICU as well as all other children requiring intensive medical treatment. A single-centre Canadian study that reviewed all deaths occurring between 2008 and 2010 found that 66% of the 220 included children died in an ICU. Of those, 53% died in an NICU and 47% in a PICU [20].

1.3.2. Circumstances of death

With respect to the high percentages of children dying in an ICU, it is not surprising that paediatric end-of-life care includes high intensity medical care and difficult decision-making. In fact, paediatric deaths in an ICU are mostly preceded by a decision to withdraw life-sustaining interventions [20]. A cross-cultural study from the US, Canada and the Netherlands confirmed in 2010 that withdrawal of artificial ventilation was the primary mode of death in neonates internationally. They found that this was the case in 69% to 93% of all NICU deaths [21]. Even higher percentages of more than 90% of primary non-intervention and redirection of care were reported in a Swiss single-centre study over a 10-year period between 1997 and 2006 [22].

Do not resuscitate (DNR) orders were reported to be in place for many children at time of death [23, 24]. Frequently, this decision was taken only shortly before the actual time of death [19, 23]. This circumstance is in accordance with the high percentages of treatment withdrawal and, as a result, only a few deaths are preceded by cardiopulmonary resuscitation (CPR). In the aforementioned Canadian study this was the case in 6% of the patients [20]; however, those numbers might be highly influenced by the type of the child's underlying life-limiting CCC. A US single-centre study conducted in children that died between 2007 and 2009 with a primary cardiac diagnosis reported that 56% of the 111 patients included underwent CPR at some point during their final hospitalisation before death [25].

1.3.3. Interventions at EOL and medications

As a consequence of the high intensity pattern of paediatric EOL care, children are exposed to invasive interventions and polypharmacy, i.e. use of several medications [13]. The most common interventions described in a US cohort of 515 children receiving PPC services at six different study sites were feeding tubes (60%), central venous catheter (22%), and tracheostomy (10%) [13]. Interventions described in the present literature vary to some degree by variables assessed, but mainly by the underlying life-limiting CCC of the child receiving EOL care. In a US/Canadian multi-site study of 275 children with progressive, non-curable genetic, metabolic, or neurologic conditions, feeding tubes were also a prevalent intervention (54%) followed by routine suctioning (18%), and oxygen administration (11%) [24]. In the aforementioned study with 111 children with a primary cardiac diagnosis 92% were mechanically ventilated and mostly sedated, 46% received mechanical circulatory support, i.e. extracorporeal membrane oxygenation (ECMO), and 23% had a peritoneal drain. The median number of surgical procedures was one per patient, with a range from zero to six [25]. A similar pattern was described in another US study conducted in two paediatric cardiology centres including 50 children who died in the hospital between 2007 and 2009. They reported that 86% of the patients were intubated, 46% needed ECMO support, 24% had gastrostomy tubes, and 22% had peritoneal drains [26].

Extensive medication profiles have been described in children receiving palliative and EOL care. Reported numbers from newer studies varied between a median of 9 different medications (*range* = 0 – 18) in Feudtner et al. [13], to a mean of 3.5 different drug categories (*range* = 0 – 12) in Steele et al. [24]. Certain drugs or groups of drugs were predominantly used, such as paracetamol, albuterol (Ventolin), antacids and anxiolytics [13, 24]. In specific populations, such as cardiological or neurological patients, inotropic agents [25, 26] or anticonvulsants were also frequently used [24].

1.3.4. Symptoms and suffering

At the centre of the highly intensive medical EOL care presented so far, there is a dying child with his/her symptoms and suffering. Suffering has been described as a state of distress that is present when the integrity of a person is disrupted. When the integrity of a child is threatened or disrupted, suffering is extended to the entire family [27].

Very few studies have looked at symptoms from the perspective of the children themselves. One notable prospective study explored determinants of symptom distress in children with advanced cancer via child self-report [28]. Common symptoms like pain, fatigue, drowsiness and irritability resulted in high levels of distress. Symptom prevalence and distress was reported as worsening during the last 12 weeks of life and pain stood out as being the most common and distressing symptom. [28].

When children cannot be asked themselves due to too young age or impaired physical condition, parents often serve as a proxy in reporting their child's symptoms and suffering. Again in children with cancer, pain was reported by the majority of parents in two US studies and one study from Germany as prevalent and associated with a great deal of suffering [29-31]. Other frequently reported symptoms by parents of children with cancer were fatigue, loss of appetite, dyspnoea [30], anxiety, constipation and nausea [31]. Besides pain, suffering was highest for dyspnoea and anxiety as reported by parents. In children with advanced heart disease at EOL and younger than two years of age, parents reported difficulty breathing and feeding, pain, irritability, and sleep disturbances as common and associated with a lot or great deal of suffering. For children older than two years of age, fatigue was most commonly reported [26].

Other studies describing symptoms in children with a life-limiting CCC at EOL used chart review as a method for collecting information. Although the quality of documentation of symptoms other than pain was reported as being variable [32], some symptoms consistently presented among different studies. The most common ones included: pain, breathing problems, irritability/agitation, lack of energy, drowsiness and seizures [23, 32]. For neonates, it has been reported that almost all newborns treated in an NICU received medication for pain and sedation [33]. However, actual descriptions of symptoms, i.e. signs, are missing from the literature. Lack of symptom documentation hindered the achievement of a US single-centre study's objective to examine symptoms exhibited by neonates at EOL [33]. In another recent international study categorizing neonatal deaths, symptoms and suffering were not addressed [21].

Where and how children die due to a life-limiting CCC or prematurity has been, to some extent, described predominantly in the USA but also internationally. For some aspects of paediatric EOL care however, descriptions were limited to certain diagnostic groups or care settings and no information at all exist for Switzerland.

1.4. The provision of PPC at the EOL

The focus of PPC is on ensuring the best possible quality of life (QOL) [18, 19]. Provision of PPC therefore has dimensions that relate to the physical, emotional and spiritual well-being of each child and family and include a mix of preventive, supportive and possibly life-prolonging interventions [1]. In 2003, the US Institute of Medicine of the National Academies released a report on improving palliative and EOL care for children and their families. They recommended that clinical practice guidelines, and institutional protocols and procedures that meet the needs of children and their families should be developed and implemented. The importance of interdisciplinary care teams was emphasized [1]. This milestone report fuelled efforts in and outside the US to create specialised PPC programmes and networks to facilitate the provision of comprehensive care. Information on the scope and characteristics of those programmes are sparsely available [14, 34]. In a recent survey among 226 US children's hospitals, 112 hospitals reported having an established PPC programme that was established in the last 10 years. A consultation model, i.e. a model in which a specialised PPC team consults with the treating team in charge of the medical paediatric subspecialty that matches the child's primary diagnosis, was the favoured model of care and applied by 88% of the 112 hospitals. The consultation scope included the entire hospital in 86 hospitals and covered the whole paediatric age span from newborns to adolescents. Home-based PC and home visits, however, were only provided in 11% and 30% of programmes [34]. The main goals of consultations as reported in an earlier study were symptom management, facilitating communication and decision-making, assisting with logistics or coordination of care, and assisting with transition to home [13].

It has been recognized that PPC should be provided in all settings where it is required, or wherever the child and family choose to be [6, 35, 36]. As home is the preferred place to be for many [37], various associations/organisations/initiatives recommend that efforts to provide home-based care should be undertaken [38]. Several issues have been described related to home-based care services for children at EOL, ranging from misconceptions among professionals and families, reimbursement issues, concerns for siblings or about the house being associated with the loss of a child, safety concerns, lack of skilled service providers, and cultural/religious beliefs [38]. Four main categories of barriers and needs in paediatric home care in Germany emerged from a qualitative study with 24 German experts in the field of PPC: "(1) specific challenges and demands in palliative care for children and adolescents, (2) lack of clear legal and financial regulations, (3) gaps in the existing care delivery/need for expansion, and (4) access to services [39, p. 4]. Information on what services are included in home-based care is scarce. A recently published study in 36 children that died while they were enrolled in an US community-based PPC programme reported on details of care they were receiving during their last six months of life. Most home visits by this multiprofessional team were provided by registered nurses, followed by case managers and paediatric nurse practitioners and included a median of 24 (*range* = 1 – 121) visits [40].

So does PPC at EOL achieve its goal of sustaining or even improving QOL by providing multiprofessional and comprehensive care, including pain/symptom management as well as psycho-social-spiritual care? There is little information available about specifically chosen outcomes such as pain and other symptoms, QOL, emergency room visits and length of stays in PICUs [41]. Efforts to evaluate PPC using (patient)/parent-reported outcomes in addition to a rather performance-oriented evaluation, i.e. characteristics of patients who received PPC, were made in the home care sector, and in the cancer population [2, 41, 42]. In the US, a retrospective survey of 60 parents who had lost a child to cancer between 2002 and 2008 was conducted to compare the symptom distress and QOL of children who received EOL care from a PPC home care programme with that of children who died without exposure to specialised PPC. The results suggested that children who were enrolled in the programme experienced better parent-reported QOL as measured for example by the amount of fun they had, by having an experience that added meaning to their lives. Symptom burden and successful pain- and symptom management was similar in both groups. Children in the programme were more likely to die at home, which was in keeping with their family's wishes [41]. In Germany, a qualitative study from 2005 with bereaved parents [31] was replicated in 2010, in order to assess whether the profound development and improvements in the field of paediatric PPC that had occurred in the meantime, have led to changes in patterns of care and symptom control of children with cancer cared for at EOL. Symptom prevalence and degree of suffering were very similar in both cohorts as reported by parents. Treatment increased for all symptoms which did not, however, lead to improved treatment success. Significantly more children received home care in the recent cohort, and fewer children died in the ICU [42]. Another German research group conducted a prospective study for which one of the goals was to evaluate whether the involvement of a specialised paediatric palliative home care team increased the effectiveness of PPC as perceived by primary caregivers. Forty families reported on their child's symptoms and QOL and their QOL and caregiver burden after involvement of the home care team compared to before. Symptom control and the child's QOL as perceived by parents, and the parents' own QOL and caregiver burden were found to have significantly improved [2].

1.5. The parental perspective of their child's EOL care

Parents facing the death of their child experience a most extreme life event, resulting in an ultimate loss and intolerable pain [43, 44]. The first and most important prerequisite for the provision of compassionate PPC and EOL care is to understand the family perspective [19].

1.5.1. Parental experiences and needs

Three recent reviews, each including qualitative and quantitative studies, summarised existing evidence related to parental experiences and needs during their child's palliative and EOL care. A total of 36 studies were reviewed (29 qualitative, 7 quantitative), many of those were part of all three reviews. The studies covered reports from bereaved parents but also from healthcare professionals. Major themes that were abstracted in all reviews in a very similar way are described as follows [45-47]:

Sincere relationships and emotional, spiritual and cultural support. The importance and existence of sincere relationships with staff that are ongoing and continual was emphasized in most studies. Desirable staff features were described as compassionate, kind and respectful. Parents wanted to be respected in their role as primary caregivers and wanted staff to listen to them. Being seen as individuals created a sense of trust, and parents were confident that their child was well cared for.

Genuine communication. Sincere communication was consistently reported as being most important for parents and families. Information should be delivered in everyday language that is easy to understand for both parents and child, as ppropriate. Parents consistently preferred that bad news about their child's diagnosis, treatment and prognosis, or news that their child was dying was conveyed openly and honestly. For many parents it was also important to receive information about what would happen when their child was dying.

Decision-making. Parents mentioned the tremendous burden involved in making certain decisions. Nevertheless, they generally appreciated being involved in decision-making and felt comfortable if the guidance, support and timing were appropriate. Concerns arose when parents felt rushed to make decisions. Several important factors were considered by parents when faced with the difficult decision to withdraw life-supporting treatment of their child, including the child's quality of life, her/his chances of getting better, or pain and discomfort.

Alleviation of suffering. The need to relieve pain and symptoms in children was naturally very important to parents. High levels of pain have been described by parents. At the same time, many believed that their child's pain was managed adequately. Commonly, parents of older children reported that they did not want the alleviation of pain or other symptoms to impair consciousness, or otherwise negatively impact the child's ability to communicate with them.

Continuity, coordination and accessibility of care. Being surrounded by and in contact with the same staff was satisfying for parents and considered to be an aspect of high-quality care. Continuity and coordination of care also resulted in less parental frustration and hyper-vigilance. Parents desired continuous and thorough documentation and reporting of the child's care plan across all levels of service providers. Home care and the possibility of transitioning in and out of hospital were also seen as a requirement for high-quality EOL care.

Bereavement support. Parents of neonates in particular appreciated it when staff helped with creating mementos, e.g. hand/footprints, locks of hair, after their child died. Some also liked to bathe and dress their child and there was a need for some private time. Some form of contact with the staff after the death of the child was enormously important and most parents felt abandoned when healthcare relationships ended abruptly.

1.8.1. Deficiencies in meeting parental needs

Several deficiencies in meeting the identified needs of parents during their child's EOL care were recurrently mentioned in the articles included in the three reviews [45-47]. First and foremost, issues related to poor communication were described. Many parents were dissatisfied with *poor communication* processes and a lack of information provided. The information deficit created by missing, inadequate and non-comprehensive provision was further fostered by the parents' inability to ask the right questions. The receipt of conflicting information from different healthcare staff resulted in confusion and stress for the parents. Problems in interacting with staff were commonly reported. These encompassed mainly disrespectful behaviour from the professionals' side and the parents' perception of not being taken seriously and being avoided. Other areas for improvement identified by parents included discrepancies between the parental positive perception of pain and symptom management and their description of symptoms, lacking availability or inflexibilities of scheduling of services, e.g. home care, clinic visits, and oversights in procedures and policies. Experiences with bereavement follow-up were reported inconsistently but families who did not receive it felt disappointed and dismissed.

One major finding across several reviewed studies warrants special emphasis. Parental experience of one single negative event related to the care their child or family received could result in profound and lasting emotional distress. This finding leads to the assumption, that quality of PPC and EOL care might influence bereavement outcomes of parents who had lost a child. Support throughout terminal care and after the loss of a child was reported to have a positive impact on long-term grieving outcomes of parents who had lost a child to cancer [48]. A similar conclusion was also drawn in a mixed study review aiming, among other things, to summarise the psychosocial impact of hospital-based bereavement services offered to bereaved parents who had lost a child [43]. It is therefore crucial to provide EOL care that meets the needs of the child and her/his family, by covering several domains and supporting the family as well as possible. This leads to the next topic covering quantitative methods to assess parental experiences and needs, and the evaluation of paediatric EOL care.

The basis for understanding parental experiences and needs has been established mainly through qualitative research. In order to move forward towards evaluation of EOL care provided, quantitative assessment of the parental perspective is needed to supplement and extend qualitative findings to a wider population leading to a higher degree of certainty.

1.6. Measurement of parental experiences and needs, and related constructs

Few researchers have attempted to quantitatively assess the parental perspective on their child's EOL care. As a result there are only a few instruments, e.g. questionnaires that try to operationalise paediatric EOL care. Reasons for this might be that paediatric EOL care has not been well conceptualised so far.

1.6.1. A possible framework

One attempt to identify quality domains for family-centred care for children with life-threatening conditions was made by The Initiative for Pediatric Palliative Care (IPPC) [49]. The IPPC was especially designed to enhance the capacity of children's hospitals and general hospitals with a paediatric unit to provide PPC in response to growing research-based recognition that the healthcare system has been failing to meet the needs of dying children and their families. Their project comprised a multiphase process, starting with needs assessment research including survey and interview data from healthcare professionals and parents who had lost a child. Additionally, a group was convened with expertise in paediatric nursing, medicine and palliative care, paediatric end-of-life care ethics, quality improvement, physician behaviour change, and medical and nursing education. This group identified six quality domains and a related set of quality indicators [50]. The quality domains are as follows: *"Holistic Care of the Child; Support of the Family Unit; Involvement of Child and Family in Communication, Decision Making, and Care Planning; Relief of Pain and Other Symptoms; Continuity of Care; and Grief and Bereavement Support"* [49, 50, p. 328]. These domains were then used to guide the development of a quality improvement tool which is now known as the "IPPC's Pediatric Palliative Care Institutional Self-Assessment Tool (ISAT)" [50, 51]. It is an institutional assessment tool to assist organisations in assessing their performance on issues related to family-centred PPC.

These quality domains were later picked up and slightly adapted in the review of Truog et al. [52] that sought to facilitate and inform future interventional studies by summarising existing evidence about EOL care in the PICU setting. They concluded that domains could be used as a framework for describing and evaluating the current status of paediatric EOL in the PICU [52].

1.6.2. Existing instruments

In 2011, Meert et al. [53] published a systematic review that aimed to identify instruments potentially useful in measuring the needs of bereaved parents whose child died in a PICU. Fifteen instruments met their selection criteria that the instrument measured needs or expierences. However, none of the instruments that met these inclusion criteria was designed to specifically assess the perspective of bereaved parents on their child's care in the PICU. Some tools were designed to assess family needs in the ICU setting (adults and paediatric) in general, some assessed experiences and needs-related constructs such as satisfaction of care or parental stressors in the NICU and PICU setting, and others were related to the assessment of parental needs of children that were hospitalised in a general paediatric unit. The authors concluded that a new specific instrument was needed to assess bereaved parents need in the PICU [53]. Consequently, the Bereaved Parent Needs Assessment (BPNA) was developed to assess parental needs and needs fulfilment around the time of their child's death in the PICU [54]. The instrument demonstrated reliability and validity and the authors suggested using the instrument to assess parents' needs and fulfilment of needs within and across PICUs rather than for individual parents [54].

Other instruments used in the paediatric setting targeted satisfaction with care. Satisfaction with care is considered a needs-related construct as it refers to the level of fulfilment of needs or expectations of an individual [55]. This assumption could be challenged however, as meeting needs does not guarantee satisfaction and unmet needs do not always result in dissatisfaction [56]. Therefore, the assessment of parental experiences and needs together with satisfaction with care should be pursued to improve care. Based on this, the 57-item (five domains) EMPATHIC-N questionnaire measuring parent satisfaction in the NICU was developed and tested. It promises to be a valid quality performance indicator for measuring the care delivered as perceived by parents, however, it has not been used with bereaved parents so far [57]. The 61-item Comprehensive Assessment of Satisfaction with Care (CASC) was originally developed in the adult cancer context but was also used to assess parent satisfaction with care after their child's death in hospital. Satisfaction with care scores were high and the authors concluded that the reporting of experiences rather than simple satisfaction measures should be incorporated [58].

This conclusion was supported by other authors, who suggested that the most important aspect of evaluating quality of care is to assess whether healthcare providers responded to the expectations and preferences of the dying patient and their families [59]. This can be achieved by asking very specific questions concerning the parents' lived experiences [60]. The lack of a comprehensive and sensitive instrument that can be used to collect meaningful information from families who have experienced the death of a child has been recognised and, recently, a report on the initial development and psychometric testing of one such instrument was published [61]. An initial 144-item version, organised in six domains, was tested in 128 bereaved mothers who had lost a child due to a life-limiting CCC. Based on this initial testing including exploratory factor analysis, the number of items could be reduced and the number of domains increased from six to ten. The domains read as follows. "Connect with families; Involve parents; Share information with parents; Share information among health professionals; Support the child; Support siblings; Structures of care; Provide care at death; Provide bereavement; and Follow-up". Overall, the instrument appears valid and applicable to EOL care in a heterogeneous setting of illnesses, ages and healthcare providers. However, testing has so far been limited to bereaved mothers [61].

Two other questionnaires are worth mentioning as they were used in the paediatric oncology palliative and EOL care setting. One was developed to serve as a guide for conducting structured interviews to mainly assess symptom experience, as perceived by parents, during the last month of life of children who died of cancer. Additional topics covered in the interviews related to the quality of care services provided on different healthcare levels [62]. Since its development, this questionnaire has been applied in a series of studies in the US and Germany [30, 31, 63]. For a Swedish population-based study involving parents who lost their child to cancer, Kreicbergs et al. [48] developed a self-administered instrument with 129 questions and a total of 365 items. The questionnaire focused on parents' self-assessment of whether they had worked through their grief and whether professional and social support facilitated this process [48].

The construct of good quality paediatric EOL care is not well operationalised no conceptual model has been developed and tested. Based on existing evidence about the parental and professional's perspective of PPC and EOL care, quality domains were identified that can serve as a framework for a quantitative assessment tool. However, few self-administered questionnaires targeting parental experiences and needs during their child's EOL care have been developed and tested.

1.7. Conducting research with bereaved parents

The question of whether research is reasonable for bereaved families has been discussed, as they are clearly vulnerable subjects. The 1978 Belmont report, which led to the Declaration of Helsinki, uses the principles of autonomy, beneficence, and justice to offer a helpful framework for addressing ethical concerns [64]. Although there may be no personal benefit, parents may still find meaning in sharing their experiences for research purposes. On the one hand, beneficence -related concerns include questions on how to minimize the burden for participating parents and what steps have to be taken to support participants in distress. On the other, justice-related issues arise when an entire population is excluded from research due to a decision not made by them.

It is remarkable that in studies with this population, the response rates were as high as 80% [48, 65]. This is confirmed by personal experiences revealing that parents who have lost a child want to tell their story. However, most studies have chosen an interval of at least 6 to 12 months, so that family members have passed the immediate stages of grief. This is in concordance with the study of Maciejewski et al. [66] who were able to show that all negative grief indicators peak within six months post loss and then start to slowly decline. Nevertheless, strategies should be considered to ensure that research with vulnerable subjects is conducted compassionately and with heightened vigilance during each step of the research process. Possible strategies have been addressed and published specifically within the field of palliative care [67, 68].

1.8. Conclusion

In the past decade, several needs assessments of paediatric palliative and EOL care have been conducted [18], providing important information, which was synthesised in this chapter 1. Many of these findings have been remarkably consistent. However, system factors and the environment in which paediatric EOL care takes place are crucial for the interpretation of those findings. In the following chapter, the Swiss strategy to promote palliative care will be shortly described which leads to the introduction of a broader research project, in which this dissertation was embedded.

1.9. References

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Chapter 2
The Swiss Setting - Rationale and Aims for the PELICAN Study and this Dissertation

Similarly to many other countries, facing a growing population in need of EOL care, the Swiss Confederation and cantons have in the last decade resolved to promote palliative care in the context of the "National Health Policy". The first "National Strategy for Palliative Care" was run from 2010 to 2012 with the goal of strengthening the availability of palliative care, improving the training of physician and nursing professionals, and better informing society. The strategy was aimed at providing impulses for improvements in EOL care. Many first measures within six sub-projects, "Care", "Financing", "Awareness", "Training", "Research", and an "Interdisciplinary sub-project", were implemented in the following three years. However, the main objective was not completely achieved [1]. In October 2012, the "National Strategy for Palliative Care 2010-2012" was extended for another three years to allow more time to complete the broad implementation of the established principles. The action plan for continuing the strategy in the years 2013 to 2015 was based on the results of the first phase [2].

Specialised palliative care was a topic emphasised in the first phase. Important interpretative documents were developed, i.e. indication criteria, overview of existing specialised offerings; however, these analyses were based on the context of palliative care for adults. This limitation was recognised and acknowledged and led to an action plan specific for paediatrics: to promote PPC in Switzerland, a thorough assessment of current practices, needs, available resources and existing services is required in order to formulate and plan further measures [2]. Following this call, the **PELICAN** study (**P**aediatric **E**nd-of-**LI**fe **CA**re **N**eeds in Switzerland) was designed and will be presented in paragraph 2.2.

2.1. State of Paediatric Palliative Care in Switzerland

As in other countries too, paediatric EOL care in Switzerland is provided in a heterogeneous setting of healthcare services. However, PPC programmes with a specialised multiprofessional team are implemented in only two of the five existing university children's hospitals and in one regional children's hospital. Apart from the five university affiliated paediatric centres, there are eight cantonal children's hospitals and several regional hospitals with a paediatric unit. Several paediatric community care organisations provide healthcare services at home. Most of them are part of the Swiss association of paediatric community care (Verband Kinder-Spitex Schweiz) and are partly subsidised by the community they practice in, with a few other free-standing private organisations. There are a few long-term institutions caring for severely cognitively and physically impaired children and adults, but no children's hospices providing PPC exist in Switzerland.

2.2. Paediatric End-of-LIfe CAre Needs in Switzerland – The PELICAN study

This dissertation is embedded in the **PELICAN** study (**P**aediatric **E**nd-of-**LI**fe **CA**re **N**eeds in Switzerland, 2012 – 2015). The overarching aims of the nationwide PELICAN study were to "provide comprehensive information and to understand the current practice of EOL care (i.e. in this study, the last 4 weeks of life prior to death) in paediatric settings in Switzerland (hospital and community care) and to explore and describe parental perspectives and the perspectives of the healthcare professionals involved. Based on the results, recommendations for best-practice during this highly vulnerable and critical phase of life not only for the affected child but also for the family will be formulated." [3, p. 1942]. The PELICAN study comprised three main studies and two sub-studies, which are described in the following, and in more detail in Chapter 3:

- PELICAN I was a retrospective chart review study aiming at describing EOL care as documented in medical charts of children (0 – 18 years) who died in the years 2011 or 2012 due to cardiac, neurological, or oncological diseases, or who died during the neonatal period.
    - PELICAN ICU, a sub-study of PELICAN I, focused on the sub-sample of children who died in an ICU describing and exploring characteristics specific to the neonatal and paediatric intensive care setting.

- PELICAN II was a mixed-methods study entailing the development and testing of an instrument to quantitatively assess the parental perspective, followed by single interviews to add further insight (sequential explanatory design).
    - PELICAN HOME, another mixed-methods study and sub-study of PELICAN I and II quantitative, focused on the sub-sample of children who spent at least three of their last four weeks of life at home. This sub-study drew on quantitative data from both main studies and single interviews were conducted (embedded design) in order to identify facilitators for and barriers to EOL care at home [4].
- PELICAN III was a qualitative study applying focus group interviews with health care professionals to explore their perspectives and needs in providing paediatric EOL care.

This dissertation comprises PELICAN I and the quantitative part of PELICAN II, including the development and testing of the Parental PELICAN Questionnaire (PaPEQu) (Chapters 4 – 6).

2.3. Research gaps and rationale for this dissertation

Parents facing the death of their child experience an unimaginably painful life event with high levels of suffering in the entire family and certainly the child affected. The family's need for compassionate professional support is high. For clinicians it is therefore imperative to have knowledge about characteristics and circumstances of childhood deaths due to a life-limiting condition or prematurity, and to understand the parental perspectives of their child's EOL care in order to provide high quality care.

Paediatric palliative and EOL care has commonly adopted the consultation model as the mode of care delivery. Providing consultation services to teams of different medical subspecialties makes the work of specialised PPC teams challenging because a high level of expertise is required. A thorough understanding of paediatric subspecialties and related knowledge of disease specific aspects of paediatric EOL are needed. This understanding should go beyond the horizon of a single hospital and take into account the heterogeneous settings where care can be provided (tertiary settings, general hospitals, paediatric primary care and in the community). Current population-based knowledge and generalisability of results are severely limited by the single site design that most studies applied. Data for Switzerland as a whole are vastly missing. Switzerland is lagging behind in the development and implementation of PPC programmes incorporating a muldidisciplinary and specialised HCT. Country-specific information is needed to initiate the next steps towards the development of a Swiss model of care for paediatric palliative and EOL care that is needs-driven and accessible for all who need it. Awareness of parental experiences and needs is a crucial prerequisite.

Existing knowledge about the parental perspective mainly originates from qualitative studies and limitations in regard to the inclusion of bereaved parents of children with certain life-limiting CCC (predominantly parents of children with cancer) are recognisable. The wide variety of underlying medical conditions leads to vastly different illness trajectories and lifespans potentially influencing what parents experience during their child's EOL care. There is little evidence as to the influence of the child's underlying diagnosis on the parental perspective. Empirical evidence is needed to base best-practise on, and this includes broadening qualitative findings with quantitative data that originating from a heterogeneous healthcare setting

A review of the literature revealed few self-administered questionnaires designed to assess the experiences and needs of parents of a dying child. At the beginning of the EPLICAN study, none of the instruments available included all of the quality domains identified by the IPPC and none were applicable to the heterogeneous field of paediatric EOL care, including children of different age groups, with different underlying illnesses and in different clinical settings. This led to the decision to develop and test an instrument that is applicable to a broad range of paediatric specialties and settings and specifically assesses parental experiences and needs during their child's EOL care.

Paediatric palliative and EOL care is based on the principle that an interprofessional team should care for patients and their families ensuring the best possible quality of life. The PELICAN study will contribute to a comprehensive understanding of EOL care in Switzerland and may be of importance for other Western countries as well. The survey instrument for assessing experiences and needs of parents who went through their child's highly burdensome EOL care phase, may also be of relevance for the evaluation of current care elsewhere.

2.4. Study aims

Given the gaps identified in the existing evidence, and following the strategy of the Federal Office of Public Health in regard to PPC by conducting a thorough needs-assessment, the following specific aims have been formulated for this dissertation:

2.4.1. PELICAN I (Chapter 4):

To ccomprehensively describe, explore and compare current practices in paediatric EOL care (for this study defined as the last four weeks of life) in four distinct diagnostic groups (cardiology, neonatology, neurology and oncology) across healthcare settings including all relevant levels of healthcare providers in Switzerland.

2.4.2. PELICAN II – Instrument development and testing (Chapter 5)

To develop and test the Parental PELICAN Questionnaire (PaPEQu), an instrument used to retrospectively assess parental experiences and needs during their child's EOL care.

2.4.3. PELICAN II – Questionnaire survey (Chapter 6)

To assess the perspectives of bereaved parents who have lost a child due to a cardiac, neurological or oncological condition or during the neonatal period in order to (1) describe specific parental experiences in relation to the underlying medical condition causing the child's death, and (2) explore differences in parental perspectives among four common medical conditions responsible for childhood death.

2.5. References

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Chapter 3

Paediatric end-of-life care needs in Switzerland: current practices, perspectives from parents and professionals. A study protocol

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3.1. Abstract

Aim. To provide comprehensive information and understanding about the current practice of end-of-life care in paediatric settings in Switzerland.
Background. In Switzerland, paediatric palliative care is usually provided by teams, who may not necessarily have specific training. There is a lack of systematic data about specific aspects of care at the end of a child's life, such as symptom management, involvement of parents in decision-making and family-centred care, and experiences and needs of parents, as well as perspectives of health care professionals.
Design. This retrospective nationwide multicentre study, **P**aediatric **End-of-Life CA**re **N**eeds in Switzerland (PELICAN), combines quantitative and qualitative methods of inquiry.
Methods. The PELICAN study consists of three observational parts, PELICAN I describes practices of end-of-life care (defined as the last four weeks of life) in the hospital and home care setting of children (0–18 years) who died in the years 2011 and 2012 due to a cardiac, neurological or oncological disease, or who died in the neonatal period. PELICAN II assesses the experiences and needs of parents during the end-of-life phase of their child. PELICAN III focuses on health care professionals and explores their perspectives concerning the provision of end-of-life care.
Conclusion. This first study across Switzerland will provide comprehensive insight into the current end-of-life care in children with distinct diagnoses as well as the perspectives of affected parents and health professionals. The results may facilitate the development and implementation of programmes for end-of-life care in children across Switzerland, building on real experiences and needs.

3.2. Summary statement

Why is this study needed?

- Paediatric end-of-life care covers particular needs during a highly vulnerable phase of life, not only for the affected child or young person but also for the whole family. Knowledge about these needs is scarce, particularly in Switzerland.
- To develop a national, needs-adapted concept for paediatric end-of-life care and services, a profound analysis of current practices, experiences and needs is a precondition.

3.3. Introduction

Children living with life-limiting conditions have always been part of the healthcare system. Mortality data for children from developed countries, such as Switzerland [1] and the USA [2] highlight two important issues relevant for investigations in the field of paediatric palliative care (PPC) and end-of-life (EOL) care. Death during infancy, particularly during the first four weeks of life, represents at least 50% of deaths in children (0–18 years) [3, 4]. Beyond the first year of life, complex chronic conditions (CCCs) – such as congenital and chromosomal disorders, neurodegenerative diseases, cancer and cardiac malformations – represent the most important group of diagnoses responsible for disease-related death in childhood [5, 6]. In childhood, the number of deaths due to diseases of the nervous system and to cardiac malformations is comparable to the number of deaths due to cancer diseases [5].

3.3.1. Background

Several studies [7-10] have investigated the needs of children and their families once the child's disease became incurable and progressive, which may also be described as the 'palliative' phase of a disease. During this phase, six domains of high-quality, family-centred care have been defined: 1) support of the family unit [11, 12]; 2) communication with the child and the family about treatment goals and plans; 3) ethics and shared decision making; 4) symptom management; 5) continuity of care; and 6) grief and bereavement support [10, 13]. PPC can address these aspects; it is typically delivered through a shared care model, where the PPC team works alongside primary attending health- and non-health-related professionals and parents [2, 14, 15]. EOL care is part of PPC and focuses on 'preparing for an anticipated death and managing the end stage of a fatal medical condition' (p.34) [12]. Communication is central to the concerns of parents and thus a principle determinant of high-quality care [16, 17]. There is evidence that professional support provided in the last four weeks of life has a positive impact on the bereavement of parents [18].

4.

Dying at home is often cited as the preferred place of death for children, associated with good quality of EOL care and a 'good death' [11, 19, 20]. In reality, paediatric home-death rates, although growing, have remained as low as 20% between 1999-2003 in the USA [21]. Neonatal intensive care units (NICUs) and paediatric intensive care units (PICUs) play a particularly important role in EOL care, as they are places where a high percentage of paediatric and neonatal patients die [22]. Nevertheless, it is important to know where and under which circumstances children die and to know the impact of characteristics of a child's death on the overall satisfaction of parents with EOL care [23]. In addition, the PPC approach, particularly at EOL, challenges health care professionals, notably those from intensive care units and physicians, by its requirement that those involved undergo a transition from a primarily curative or at least life-prolonging treatment to a treatment that takes the quality of life as its main focus and anticipates the death of the child [24-26].

As a matter of fact, evidence on how to provide optimal PPC, including EOL care, which covers the needs of children and their families is scarce and lags substantially behind that in the adult population. This is particularly true in Switzerland, where there are no systematic and comprehensive records of data about the current provision of PPC and EOL care in paediatric settings. Furthermore, in contrast to many European countries, Swiss PPC and EOL care is not provided in facilities such as hospices for children. As a consequence, the question needs to be raised whether new models of care for PPC and EOL care are needed.

3.4. The study

3.4.1. Aims

The general aims of this study are to provide comprehensive information and understanding about the current practice of EOL care (i.e. in this study, the last 4 weeks of life prior to death) in paediatric settings in Switzerland (hospital and community care) and to explore and describe parental perspectives and the perspectives of the health care professionals involved. Based on the results, recommendations for best-practice during this highly vulnerable and critical phase of life not only for the affected child but also for the family will be formulated. The PELICAN-objectives of the three distinct PELICAN parts and two sub-studies are described as follows:

- **PELICAN I** aims at describing the EOL phase and EOL care as documented in medical charts of children 0–18 years of age who died in the years 2011 and 2012 due to cardiac, neurological, or oncological diseases, or who died during the neonatal period and at exploring differences between these distinct groups of diagnoses related to disease-specific aspects and settings of care.
- **PELICAN II** entails the development, piloting and preliminary psychometric testing of a survey instrument, followed by the quantitative and qualitative enquiry into parents' experiences and needs during the EOL care of their child that meets the inclusion criteria of PELICAN I.
- **PELICAN HOME**, a sub-study of PELICAN I and II that focuses on experiences and needs of parents whose child spent his or her EOL predominantly at home (≥ 21 days of the last 4 weeks of life) and thus received EOL care in the home care setting [27]. This sub-study aims at identifying facilitators and barriers regarding the provision of EOL care and at comparing results between the main settings of care in Switzerland – in hospital and in the child's home.
- **PELICAN ICU**, a sub-study of PELICAN I focuses on EOL practices specific to the population cared for in neonatal and paediatric intensive care units, compared with practices in other settings, such as general wards.
- **PELICAN III** explores perspectives and needs of health care professionals who are involved in paediatric EOL care, but not necessarily specialised in palliative care.

3.4.2. Design

This retrospective, multicentre study entails different designs for each element of the study under investigation, which are listed below and illustrated in Figure 1.

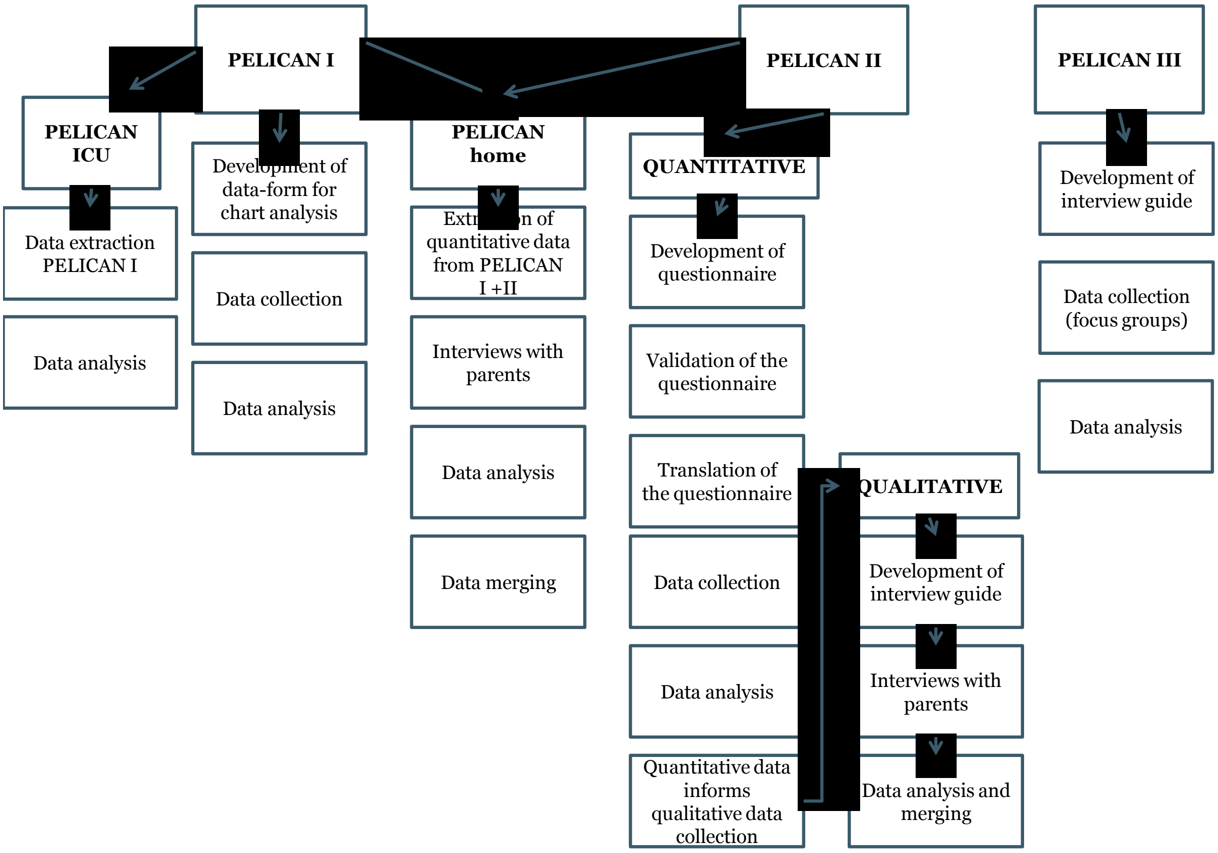

Figure 1 Study overview

Quantitative and qualitative methodology is used and combined during analyses, to explore the child's last four weeks of life from different perspectives:

PELICAN I includes the quantitative, retrospective analysis of medical charts of children (0–18 years) who died from cardiac, neurological and oncological diseases as well as during the neonatal period in the years 2011 and 2012.

PELICAN II includes a) the questionnaire development in German (item generation, content validity testing by an expert panel and through cognitive debriefing), standard translation of the questionnaire into French and Italian, pilot testing of the instrument and preliminary homogeneity testing of items and application of the questionnaire for the main survey with parents; b) face-to-face interviews with parents. To obtain a comprehensive and deeper understanding of parents' perspectives a sequential explanatory mixed methods design will be used (QUANT → qual). The combination of quantitative and qualitative enquiry methods will enhance analyses by exploring this existential human experience in a cultural and social context [28, 29].

PELICAN HOME is a mixed-method sub-study of PELICAN I and II that focuses on EOL care in the home care setting [27].

PELICAN ICU is a retrospective descriptive sub-study of PELICAN I and focuses on EOL care in the paediatric and neonatal intensive care setting.

PELICAN III is a qualitative study using five to six interdisciplinary focus group interviews entailing around 48 health care professionals that will be performed across Switzerland in tertiary and secondary level hospitals.

3.4.3. Setting

Sixteen hospitals, two long-term institutions and ten community care organizations across the entire country have agreed to participate in the study. In view of the three language regions of Switzerland, close collaboration has been developed with the French and Italian speaking parts by designating local teams who will take over responsibility for the coordination of the study in their region.

3.4.4. Participants

The recruitment of subjects for PELICAN I and II was completed in the time period from August 2013 - April 2014. To identify subjects and participants, all children's hospitals or hospitals with a paediatric ward and the paediatric community care organisations in Switzerland were contacted between August and December 2013.

Inclusion criteria for children were age at the time of death (0–18 years, but not younger than 24 hours) and diagnostic group (cardiology, neonatology, neurology, oncology) even if a child died due to a complication, such as recurrent pneumonia and the underlying disease was a neurological disorder. Subjects were excluded if a child: died within 4 weeks following a traumatic injury; died within 2 days after the diagnosis of an oncological disease; died unexpectedly due to a treatment or procedural medical error; or if his or her family were not Swiss residents or did not speak/read German, French or Italian.

Parents whose child met the eligibility criteria were mailed an invitation letter by the former treating hospital and asked to fill out an informed consent document accepting or declining study participation (i.e., allowing the use of data from their child's medical charts, participation in the questionnaire survey and/or agreeing to be contacted for an interview). Return of the completed questionnaire was considered to be formal agreement to the analysis of the survey data. Additional written informed consent will be obtained from parents who participate in the interviews. Participants for the qualitative part of the PELICAN HOME sub-study have been selected in the German-speaking part of Switzerland only. For the overall PELICAN project, we expected a total of approximately 380-400 eligible patients and a response rate of 60%.

For PELICAN III, health care professionals (paediatricians, paediatric nurses including community nurses) and allied-health professionals (psychologists, social workers, physiotherapists and other therapists and pastors) working in participating hospitals or paediatric community care will be invited to take part in 5 to 6 focus group interviews with approximately 8 participants in each focus group. Three of these group meetings will be performed in the German-speaking part of Switzerland, one to two in the French- and one in the Italian-speaking part.

3.4.5. Data collection

For PELICAN I, we developed a secure web-based data-form (secuTrial®) with approximately 200 variables to facilitate data entry and management. These variables were selected according to the literature and our study objectives. They include data to verify eligibility, patients' characteristics (e.g. demographic data, diagnosis), characteristics and circumstances of death (place of death and life-sustaining treatment during the last 24 hours of life), treatment modalities (palliative care approach, interventions requiring anaesthesia, medical devices, type and dosage of drugs, nutrition and fluids regimens), treatment modalities specifically related to cancer diagnoses, EOL care in particular (e.g. support of the family unit, communication with the child and the family, decision making, pain and symptom management, continuity and coordination of care) and grief and bereavement support. The data-form was pilot tested with ten children who died in 2010. Only minor adaptations had to be made and the accessibility of data from medical charts was evaluated as sufficient. Data collection from patients' medical charts started in November 2013 and was completed in June 2014. At each participating study site (hospital, community care centre) local coordinators have been assigned to facilitate logistics concerning medical charts review and workspaces.

For PELICAN II, a survey instrument was developed, as no instrument that met the criteria for our study objectives was found. Based on the quality domains for family-centred EOL care the questionnaire was structured into six themes: 1) Support of the family unit; 2) Communication with the child and family; 3) Ethics and shared decision making; 4) Relief of pain and other symptoms; 5) Continuity of care; and 6) Grief and bereavement support. For each of the four groups of diagnoses (cardiology, neonatology, neurology and oncology), separate versions were developed and designed for mothers and fathers. Each questionnaire comprises approximately 100 items (including socio-demographic factors and questions related to overall parental satisfaction). Questions are predominantly closed-ended and are presented in various formats, yet the majority uses Likert-type or categorical response options. Validity testing included face validity and content validity indexing with 26 experts and cognitive debriefing with 4 affected parents.

A purposefully selected sample of approximately 20 parents who completed the questionnaire and also consented to participate in an interview will be contacted by phone for an appointment for the interview. The interviews will be conducted by a trained study collaborator at the parents' home or a place of their preference, audiotaped and transcribed verbatim. They will last approximately 60 minutes and will be conducted from January to April 2015. An interview guideline will be developed according to the results from PELICAN I and II.

For PELICAN III, an interview guideline for the focus group interviews will be developed that reflects the current status of care and takes into consideration potential barriers and facilitators for the provision of child- and family-centred care, PPC and EOL care. In addition, experiences and needs of health care professionals will be explored.

3.4.6. Data analysis

Quantitative data analysis

All data from PELICAN I and II will undergo an exploratory analysis to describe the data and uncover any data inconsistencies that may impact the validity of the data analysis. Appropriate descriptive statistics will be used to summarise data of individual variables, considering each variable's level of measurement and its observed distribution (e.g. frequency distributions with counts and percentages, measures of central tendency and dispersion). To test for differences between diagnostic groups, clustering effects of parents of the same child will be accounted for by using multilevel approaches. Data will be analysed using SPSS Statistics® version 21 for Macintosh (SPSS Inc., Chicago, IL, USA) with P-values of <0.05 considered significant.

Qualitative data analysis

Parental and also focus group interviews will be analysed by the method of 'thematic analysis' [30]. Thematic analysis is an analytic method for identifying, analysing and reporting patterns of themes in a data set. Themes will be placed into a semantic context and further examined in relation to frameworks defined by us for each part of qualitative data enquiry (e.g. Feudtner *et al.*'s multi-level system model for the analysis of parents' interviews in the PELICAN HOME sub-study [6, 31]). After systematic analysis of each interview, the latter step will take place in a research team to deepen our understanding of the particularities of each perspective and to reach high congruence, credibility and validity of the results. ATLAS.ti® version 7 for Microsoft Windows® (ATLAS.ti GmbH, Berlin) will support data management and organisation.

Mixed methods data analysis

After sequential data collection and separate analyses, the quantitative data results will be compared with major themes identified across the qualitative data set using side-by-side comparison on a summary table. Results of this comparison will be discussed in the context of developed frameworks and system models. Mixed methods analysis will provide a comprehensive picture and a fuller understanding of the different perspectives, experiences and needs.

3.4.7. Ethical considerations

Research with bereaved parents may provoke acute grief reaction and may thus be of particular burden for participants. However, studies with these subjects have shown high response rates, as high as 70-80% [32-35], which can be interpreted in the way that parents appreciated to share their experiences and burden and benefit are rather balanced. Nevertheless, we decided to ensure a timeframe of at least one year between the child's death and participation in the study as this time has been found to be the most acute and vulnerable [36]. In addition, participation in this study is entirely voluntary. Eligible participants are invited with written information through the treating centre. Written informed consent is a precondition for all three parts of the study. Ethics committee approval has been obtained by the leading Human Research Ethics Committee (March 2013) and ten departmental committees across Switzerland.

3.4.8. Validity and reliability

For PELICAN I, the data-form was pilot tested with ten children who died in 2010. Only minor adaptations had to be made and the accessibility of data from medical charts was evaluated as sufficient. In the main study, 5% of the medical charts will be randomly chosen and two different persons will enter the data independently for quality checking.

For PELICAN II, the German version of the questionnaire underwent content validity testing by an expert panel and through cognitive debriefing with parents. Following standard translation into French and Italian, a pilot testing of the instrument and preliminary homogeneity testing of items and application of the questionnaire for the main survey with parents was performed. Qualitative data from interviews with parents and focus groups (PELICAN II and III) will be analysed systematically according to Braun and Clarke [30] and discussed in research teams (5-6 participants) to ensure accuracy of the analytical process and to reach high congruence, credibility and validity of the results.

3.5. Discussion

When the burden of a life-limiting disease increases and poor quality of life outweighs the potential benefit of life prolongation, the focus of care shifts towards individual needs of the child and his or her family. Preferences of the child and the family move into a focus of care that may be associated with difficult decisions about continuation or discontinuation of life-sustaining therapies, anticipation of the process of dying, pain and symptom management and planning of the location and/or circumstances of death. Even though PPC has made great advances during the last two decades, many needs of children and their families remain unmet [18, 37-40] and several of them are probably unknown. This study covering neonatal and paediatric EOL care will contribute to a comprehensive understanding of EOL care in Switzerland and may be of importance for other Western countries as well. The survey instrument for assessing the experiences and needs of parents who went through their child's highly burdensome palliative and EOL care phase, may be of relevance for the evaluation of current care elsewhere; the impact of innovations in a distinct institution on the experience of parents may likewise be of broader interest.

3.5.1. Limitations

The current study has several limitations. First, data quality of the retrospective chart analysis will be moderate due to the study design with probably sparse documentation in hospitals and community care organisations. Second, due to the small size of the country and different language regions, we might gain a rather heterogeneous picture, which might hamper generalisation of our results, even for Switzerland. Third, it explores the situation in Switzerland and may thus be of limited relevance for other countries.

In conclusion, this survey across Switzerland will provide first comprehensive insight into the current practice of EOL care in children suffering from the four largest and most important circumstances of disease-related death in childhood.

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3.8. Conflict of interest

No conflict of interest has been declared by the authors.

3.9. Author contributions

All authors have agreed on the final version and meet at least one of the following criteria [recommended by the ICMJE (http://www.icmje.org/ethical_1author.html)]:

- substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data;
- drafting the article or revising it critically for important intellectual content.

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Chapter 4

Patterns of care at end-of-life in neonates and children with complex chronic conditions: A nationwide chart review among different care settings

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4.1. Abstract

Background: End-of-life care of neonates, children and adolescents is challenging and requires a high level of professional expertise. Specialised paediatric palliative care has commonly adopted a consultation care model. It is important that teams have a thorough understanding of paediatric subspecialties and related knowledge of disease specific aspects of paediatric end-of-life care.
Aim: To comprehensively describe, explore and compare current practices in paediatric end-of-life care in four distinct diagnostic groups including all nationwide relevant levels of healthcare providers.
Design: Nationwide retrospective chart review.
Setting/participants: Data from 149 paediatric patients who died in the years 2011 or 2012 due to a cardiac, neurological or oncological condition, or during the neonatal period were collected in 13 hospitals, two long-term institutions and 10 community-based healthcare service providers throughout Switzerland.
Results: Sixty-two percent of the patients died in intensive care units, 84% of them following the withdrawal of life-sustaining treatment. A smaller proportion (17%) died at home. Reliance on invasive medical interventions was prevalent, and the use of medication was high, with a median count of 12 different drugs during the last week of life. Patients experienced an average number of 6.42 symptoms. Symptom count was significantly lower (M = 4.39) in neonates and the prevalence of various types of symptoms differed significantly among the four diagnostic groups. Only half of the patients who spent some days at home received community-based healthcare.
Conclusions: The study provides a comprehensive overview of current end-of-life care practices in the heterogeneous real-life setting of hospitals, long-term institutions and community healthcare organisations in Switzerland. The findings provide a knowledge base for paediatric palliative care teams. As a consequence, a meaningful outcomes measure should be introduced into practice to promote quality of care and further advance research in the field.

4.2. Key statements

What is already known about the topic?

- Various medical diagnoses contribute to childhood deaths due to complex chronic conditions encompassing the whole paediatric age span.
- Characteristics and circumstances of end-of-life care are known to vary by age and medical condition.
- No study has so far focused on comparisons between distinct diagnostic groups.

What this paper adds?

- Differences in characteristics and circumstances of end-of-life care are present, depending on the underlying medical condition.
- Professional home care is established for a few patients only.
- The types of community healthcare services are manifold.

Implications for practice, theory or policy?

- As the field of paediatric palliative care is growing internationally, specialists need comprehensive knowledge to base their practice on.
- The capacity of community healthcare services needs to be expanded.
- Bridging the gap among service providers should be emphasized.

4.3. Introduction

Despite continued advancements in medical care and improved survival or life expectancy, childhood deaths due to complex chronic conditions (CCC) or prematurity are inevitable [1]. Deaths during the first year of life constitute approximately 50% of disease-related deaths, the causes of which include perinatal complications, prematurity, or congenital anomalies [2-4]. Beyond the age of one year, the three most common life-limiting CCCs are neurological/neuromuscular and cardiovascular conditions (including genetic disorders), and malignancies [1, 2, 5]. The majority of disease- and prematurity-related deaths occur in hospitals,[6-8] and for children dying at home, hospital use in their terminal stage is high [1, 2]. Symptom burden and reliance on medical technology has been reported to be considerable [9, 10]. Circumstances and characteristics of deaths, however, are known to vary by age and medical conditions [1, 2]. Care is normally planned and performed by the team of the paediatric medical subspecialty matching the child's diagnosis.

Paediatric palliative (PPC) and end-of-life (EOL) care emerged as a medical subspecialty aimed at meeting the specific needs of seriously ill children and their families. Meeting these needs requires a comprehensive and integrative approach from a compassionate and skilled multidisciplinary team [11]. PPC should be provided in all settings where it is required [12], although, specialised PPC teams are mostly hospital based [13]. In a recent survey among 226 US children's hospitals, the majority of the 112 hospitals with an established PPC programme reported that they offered consultation services throughout the hospital and that their programme covered the whole paediatric age span from newborns to adolescents [13].

Providing consultation services to teams of different medical subspecialties makes the work of specialised PPC teams challenging because a high level of expertise is required. A thorough understanding of paediatric subspecialties and related knowledge of disease specific aspects of paediatric EOL are needed. This understanding should go beyond the horizon of a single hospital and take into account the heterogeneous settings where care can be provided (tertiary settings, general hospitals, paediatric primary care and in the community). There is not much evidence on which to base best practice and most existing studies focus on specific diagnostic cohorts or specific care settings [1, 14, 15]. It was therefore the aim of this national study to comprehensively describe, explore and compare current practices in paediatric EOL care (for this study defined as the last four weeks of life) in four distinct diagnostic groups (cardiology, neonatology, neurology and oncology) across healthcare settings including all relevant levels of healthcare providers in Switzerland.

4.4. Methods

4.4.1. Study design

This retrospective chart review was part of PELICAN (Paediatric End-of-LIfe-CAre Needs in Switzerland, 2012-2015, NCT01983852), a nationwide study "to provide comprehensive information and to understand the current practice of EOL care (i.e. in this study, the last 4 weeks of life prior to death) in paediatric settings in Switzerland (hospital and community care) and to explore and describe parental perspectives and the perspectives of the healthcare professionals involved" [16]. Human Research Ethics Committees from the 11 Swiss cantons in which the study took place approved the PELICAN study (leading committee: KEK ZH Nr. 2012-0537). Parents who had lost a child due to a cardiac, neurological or oncological condition or during the neonatal period (independent of the underlying condition) in the years 2011 and 2012 were invited to participate. Neonates < 24 hours of life and patients > 18 years were excluded. Information on how, where and when recruitment took place is described in detail elsewhere [17].

4.4.2. Setting and data collection

Data from all eligible patients, whose parents had consented to the review of their child's medical chart, were collected in 13 hospitals, two long-term institutions and 10 community-based healthcare service providers throughout Switzerland. Among the 13 hospitals, there were five tertiary paediatric centres, four dedicated children's hospitals, three general hospitals with paediatric units and one tertiary care centre with a neonatal intensive care unit. A multiprofessional PPC team was available in two paediatric centres and one children's hospital; no paediatric hospices exist in Switzerland.

Data collection took place between November 2013 and June 2014. It was conducted mainly by the first author, who also developed the coding manual, all case report forms and instructed and supervised five assistants, who supported data collection [18]. In accordance with this study's definition of EOL care being care during the last four weeks of life, data collection was restricted to the 28 days prior to the child's death. All extracted data was entered into secuTrial®, a browser-based electronic data capture system (InterActive Systems, Berlin, Germany). Five percent of the medical records reviewed by an assistant were promptly audited by the first author to detect and correct discrepancies [18]. Inconsistencies and emerging questions were continuously discussed among data collectors to ensure the quality of ongoing data extraction and reduce the likelihood of inter-rater discrepancies.

4.4.3. Variables

The following data were collected for this study: (1) demographics (age, gender); (2) diagnostic information (the underlying diagnosis primarily responsible for the patient's death, gestational age for newborns only, time since diagnosis, and whether the diagnosis was made prenatally); (3) circumstances of death (place of death, occurrence of resuscitation, existence of do not resuscitate [DNR] orders and whether these orders changed during the last four weeks of life, and treatment withdrawal); (4) interventions and medications (anaesthesia, ventilation, central access device, enteral feeds, number and types of medications); (5) symptoms (presence of various symptoms); (6) hospital and community healthcare utilisation (hospital days and admissions, days spent at home, number of days and hours, and types of care provided by community services).

A diagnostic chapter and code from the International Statistical Classification of Diseases and Related Health Problems (ICD), 10th Revision, online version 2016 [19] was assigned by two investigators to each patient, based on the exact diagnostic information documented in the patient's last medical report. All symptoms documented in the patient's chart were recorded during data collection. The ones most frequently reported were categorised into 20 common symptoms based on existing literature [10, 14, 20]. Symptoms that affected similar areas like spasticity/dystonia for muscular impairments, or agitation/irritability for behavioural problems were grouped.

4.4.4. Statistical analysis

Descriptive statistics (measures of central tendency and dispersion, frequencies and percentages) were used to explore and summarize all variables. A binary logistic model with likelihood ratio statistics was utilised for two-tailed comparisons between the diagnostic groups of variables with a binominal response (Yes – No). For count outcome variables, negative binomial regression was utilised to adjust for overdispersion [21]. The neonatology group was specified as reference category, as it was the largest group, with results that often differed considerably compared to the other groups. For variables with a categorical response, equivalence of proportions between diagnostic groups was tested in contingency tables using the Pearson's chi-square test or Fisher's exact test when cell sizes were < 5. Because missing data were rare (reported in the tables) we did not pursue measures for missing value replacement. Due to the multiple comparisons performed, we set a conservative p-value of > 0.001 to indicate significance. Statistical analyses were performed using IBM© SPSS© Statistics 21 for Mac® (IBM Corp, Armonk, NY, USA).

4.5. Results

Of the 307 eligible families, 267 could be contacted and were invited to participate in the PELICAN study. Of those, 147 families (55%) consented. Two families lost twins resulting in a study sample of 149 neonates, children and adolescents (Table I). With neonates comprising 38% of the sample, the median age at death was 0.5 years for the entire sample but substantially higher (*Mdn* = 8 years) for the oncology group. Seven ICD-10 diagnostic chapters were represented in our four groups' categorisation, with the highest variety found within the neurology group. The median time between diagnosis of the life-limiting CCC and death for the total sample was one month (*interquartile range [IQR],* 0 – 6). Within the four groups, the median time between diagnosis and death was longest for the neurology group (*Mdn* = 6 months, *IQR* = 3 – 29). Diagnoses made prenatally, which were not taken into account in the above calculation, were significantly more frequent in the cardiology group compared to the other groups (p = < 0.001) and not present in the oncology group.

Table I. Demographic and diagnostic patient characteristics

Characteristics	**Total** *N* = 149 (100%)	**Cardiology** *n* = 19 (13%)	**Neonatology** *n* = 57 (38%)	**Neurology** *n* = 36 (24%)	**Oncology** *n* = 37 (25%)
Age, *Mdn* (*range*)					
in days	Na	Na	5 (1 – 26)	Na	Na
in months	6 (0 – 209)	6 (1 – 109)	Na	19 (1 – 207)	101 (20 – 209)
in years	0.5 (0.0 – 17.4)	0.5 (0.1 – 9.1)	Na	1.6 (0.1 – 17.2)	8.4 (1.7 – 17.4)
Gender, *n* (*%*)					
Female	72 (48)	10 (53)	32 (56)	15 (42)	15 (40)
Male	77 (52)	9 (47)	25 (44)	21 (58)	22 (60)
ICD-10 chapter, description, *n* (*%*)					
II Neoplasms	36 (24)	0 (0)	0 (0)	0 (0)	36 (97)
III Blood/immune system	1 (1)	0 (0)	0 (0)	0 (0)	1 (3)
IV Endocrine, nutritional, metabolic	6 (4)	0 (0)	0 (0)	6 (16)	0 (0)
VI Nervous system	21 (14)	0 (0)	2 (4)	19 (53)	0 (0)
IX Circulatory system[a]	5 (3)	4 (21)	0 (0)	1 (3)	0 (0)
XVI Conditions originating in perinatal period	45 (30)	0 (0)	44 (77)	1 (3)	0 (0)
XVII Congenital, chromosomal	35 (24)	15 (79)	11 (19)	9 (25)	0 (0)
Gestational age (for the neonatology group only)			***n* = 52**[b]		
24 0/7 – 27 6/7	Na	Na	17 (33)	Na	Na
28 0/7 – 31 6/7	Na	Na	8 (15)	Na	Na
32 0/7 – 36 6/7	Na	Na	9 (17)	Na	Na
37 0/7 - > 42 0/7	Na	Na	18 (35)	Na	Na
Time since diagnosis[c]	***n* = 139**[c]	***n* = 18**[c]		***n* = 28**[c]	***n* = 36**[c]
in days, *Mdn* (*range*)	Na	Na	4 (1 – 26)	Na	Na
in months, *Mdn* (*range*)	1 (0 – 167)	6 (0 - 66)	Na	6 (0 - 167)	4 (0 - 139)
in years, *Mdn* (*range*)	0 (0 – 14)	0.5 (0.0 – 5.5)	Na	0.5 (0.0 – 14.0)	0.5 (0.0 – 12.0)
Diagnosis made prenatally	***n* = 139**[c]			***n* = 34**[c]	
Yes, *n* (*%*)	31 (21)	11 (58)	13 (23)	7 (21)	0 (0)

Note. Na = Not applicable. ICD-10 = International Classification of Diseases, 10th Revision.

[a] Stroke included. [b] Information was missing for some cases. [c] Calculated from date of birth, even if diagnosis was suspected prenatally.

Table II. Place and circumstances of death

	Total *N* = 149 (100%)	**Cardiology** *n* = 19 (13%)	**Neonatology** *n* = 57 (38%)	**Neurology** *n* = 36 (24%)	**Oncology** *n* = 37 (25%)	***p*-value**
Place of death, *n* (%)						< 0.001[a]
PICU	63 (42)	13 (67)	27 (48)	13 (36)	10 (27)	
NICU	30 (20)	0 (0)	27 (48)	3 (8)	0 (0)	
Hospital / long-term institution	26 (18)	2 (11)	0 (0)	13 (36)	11 (30)	
Home	25 (17)	2 (11)	2 (3)	7 (20)	14 (38)	
Emergency department / Transport	5 (3)	2 (11)	1 (1)	0 (0)	2 (5)	
CPR[b]						
Yes, *n* (%)	26 (17)	7 (37)	6 (11)	7 (19)	6 (16)	0.097[c]
DNR order					***n* = 35[d]**	
Yes, *n* (%)	91 (62)	11 (58)	20 (35)	33 (92)	27 (77)	< 0.001[c]
DNR status change within the last four weeks of life	***n* = 147[d]**			***n* = 35[d]**	***n* = 36[c]**	
Yes, *n* (%)	93 (63)	11 (58)	50 (88)	16 (46)	16 (44)	< 0.001[c]
Withdrawal of life-sustaining interventions[e]	***n* = 93**	***n* = 13**	***n* = 54**	***n* = 16**	***n* = 10**	
Yes, *n* (%)	78 (84)	10 (77)	49 (91)	12 (75)	7 (70)	0.203[c]

Note. PICU = Paediatric intensive care unit. NICU = Neonatal intensive care unit. CPR = Cardiopulmonary resuscitation. DNR = Do not resuscitate.
[a]Fisher's exact test. [b]Within 24 hours before death. [c]Likelihood ratio chi-square. [d]Information was missing for some cases. [e]Only applies to patients who died in an intensive care unit.

4.5.1. Place and circumstances of death

Ninety-three patients (62%) died in an intensive care unit (ICU) with the highest proportion (96%) of these patients being in the neonatology group (Table II). Twenty-five patients (17%) died at home with the highest proportion (38%) in the oncology group. Twenty-six patients (17%) received cardiopulmonary resuscitation (CPR) within 24 hours before death. A do not resuscitate (DNR) order was documented in 91 patients' charts (62%). The DNR status changed within the last four weeks of life for 93 patients (63%), most often within the neonatology group (*n* = 50, 88%) and shortly before the child's death. For 78 patients (84%) of the 93 who died in an ICU, death was preceded by a decision to withdraw life-sustaining interventions.

4.5.2. Interventions, medication and symptoms

During the last four weeks of life, patients underwent several interventions, suffered from a variety of symptoms, and received a considerable amount of medication as documented in their charts. This information is detailed in Table III and Figure 1. The most common documented interventions requiring anaesthesia were surgical interventions in 28 patients (55% of the 51 patients that received anaesthesia) and diagnostic procedures, e.g. imaging in 27 patients (53%). The number of medications with orders for standard daily doses and as needed orders was assessed for the last two weeks of life. The overall median number rose from 9 (*range* = 0 - 42) during the second last week to 12 (*range* = 1 – 46) during the last week. For 133 patients (89%) the last treatment approach was documented as palliative. However, the approach changed during the last month in 88 patients (59%), most commonly in the neonatology group and least commonly in the oncology group (90% vs. 32%, $p = < 0.001$).

Pain was the most frequent symptom documented, and occurred in 110 patients (78%, N = 141) with no significant differences between the diagnostic groups. One hundred and forty patients (95%, N = 148) received some sort of pain medication, most commonly opioids (93%) followed by paracetamol (67%), the latter predominantly in oncology patients. Other common symptoms included breathing problems (n = 107, 72%), followed by behavioural problems such as agitation or irritability (n = 89, 60%). Some symptoms such as respiratory secretion, fever, nausea/vomiting, coughing, sweating, fatigue, drowsiness, anxiety, including worry and sadness, and poor appetite differed significantly ($p = < 0.001$) between the diagnostic groups (Figure 1). Overall, an average of 6.42 (*SD* = 3.14) symptoms were recorded per patient. Significantly fewer symptoms were reported in neonates (*M* = 4.39, *SD* = 2.15) compared to all other groups ($p = < 0.001$).

4.5.3. Hospital and community healthcare utilisation

Overall, our study patients stayed in the hospital for a median of six days (*IQR* = 2 – 19) during their last four weeks of life, with the highest number of hospital days for patients in the cardiology group (Table IV). Twenty patients (13%) had no hospital days, 12 (60%) from the oncology, 5 (25%) from the neurology, 3 (15%) from the cardiology, and zero from the neonatology group. Among the 129 patients who had at least one hospital day,61 patients (47%) had one hospital admission, 11 patients (8.5%) two, and 2 patients (2%) had 3 admissions during the last four weeks of life. Fifty-six patients (43%) had zero hospital admissions, meaning that those patients were hospitalised at the beginning of data collection and remained there until their death or discharge. Of the 57 patients in the neonatology group, 23 patients (40%) were born in a hospital with no ICU and had to be transferred to a referral tertiary hospital with an ICU. Patients from the other diagnostic groups were most commonly admitted from home (Table IV).

Seventy-two patients (48%) stayed at home for at least one day during their last four weeks of life, with patients from the oncology group having the highest number of home days (*Mdn* = 24, *IQR* = 4 - 28), followed by patients from the neurology group (*Mdn* = 21, *IQR* = 4 - 26). Of the 72 patients who stayed at home, 36 (50%) received professional care from a community-based service. The provision of education and support to empower the family was the most commonly provided service as documented by the care provider, and patients from the neurology group received more care hours than patients from the other groups (Table IV).

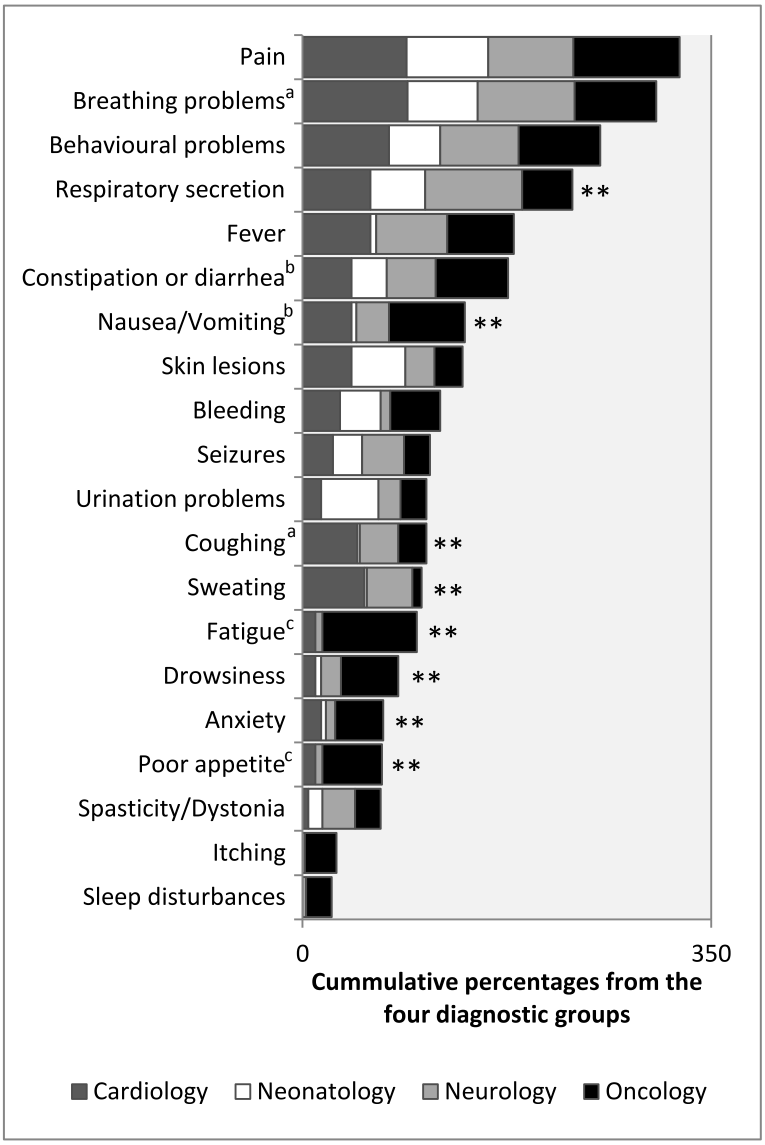

Figure 1. Symptom prevalence and comparison between the four diagnostic groups.
Note. ** = *p*-value < 0.001.
[a]Adjusted for mechanical ventilation. [b]Adjusted for enteral feeds.
[c]Neonatology group excluded due to 0 % of symptom presence.

Table III. Interventions and medications during the last four weeks of life

	Total *N* = 149 (100%)	**Cardiology** *n* = 19 (13%)	**Neonatology** *n* = 57 (38%)	**Neurology** *n* = 36 (24%)	**Oncology** *n* = 37 (25%)	***p*-value**
Interventions requiring anaesthesia				***n* = 35**[a]		
Yes, *n (%)*	51 (35)	11 (58)	21 (37)	6 (17)	13 (35)	0.021[b]
Mechanical ventilation						
Yes, *n (%)*	94 (63)	14 (74)	55 (97)	15 (42)	10 (27)	< 0.001[b]
ECMO						
Yes, *n (%)*	7 (5)	4 (21)	3 (5)	0 (0)	0 (0)	< 0.001[b]
CAD	***n* = 148**[a]				***n* = 36**[a]	
Yes, *n (%)*	106 (72)	14 (74)	55 (97)	12 (33)	25 (69)	< 0.001[b]
Enteral feeds via nasogastric or gastrostomy tube	***n* = 148**[a]			***n* = 35**[a]		
Yes, *n (%)*	114 (77)	17 (90)	51 (90)	33 (94)	13 (35)	< 0.001[b]
Medication count in the last week of life[c]	***n* = 146**[a]			***n* = 35**[a]	***n* = 35**[a]	
Mdn, (range)	12 (1 - 46)	19 (3 - 45)	12 (1 - 34)	10 (3 - 39)	13 (4 - 46)	0.006[b]
Pain medication	***n* = 148**[a]			***n* = 35**[a]		
Yes, *n (%)*	140 (95)	18 (95)	54 (95)	33 (94)	35 (95)	1.000[b]
Anxiolytic medication	***n* = 145**[a]			***n* = 35**[a]	***n* = 34**[a]	
Yes, *n (%)*	84 (58)	14 (74)	27 (47)	18 (51)	25 (74)	0.032[b]
Antiemetic medication	***n* = 146**[a]			***n* = 35**[a]	***n* = 35**[a]	
Yes, *n (%)*	25 (17)	2 (11)	0 (0)	0 (0)	23 (66)	< 0.001[b]

Note. ECMO = Extracorporeal membrane oxygenation. CAD = Central access device, either venous or arterial.
[a]Information was missing for some cases. [b]Likelihood ratio chi-square. [c]Includes both standing daily dosages and as-needed orders.

Table IV. Hospital and community healthcare utilisation during the last four weeks of life

	Total *N* = 149 (100%)	**Cardiology** *n* = 19 (13%)	**Neonatology** *n* = 57 (38%)	**Neurology** *n* = 36 (24%)	**Oncology** *n* = 37 (25%)	***p*-value**
Hospital days, *Mdn* (*range*)	6 (0 - 28)	20 (0 - 28)	5 (1 - 26)	7 (0 - 28)	4 (0 - 28)	0.035[a]
Care setting before hospital admission[b], *n* (*%*)	***n* = 88[c]**	***n* = 17**	***n* = 23**	***n* = 26**	***n* = 22**	
Home	37 (42)	11 (64)	0 (0)	16 (62)	10 (45)	NA[d]
Other hospital	35 (40)	3 (18)	23 (100)	4 (15)	5 (23)	NA[d]
Emergency department	8 (9)	3 (18)	0 (0)	4 (15)	1 (5)	NA[d]
Outpatient clinic	7 (8)	0 (0)	0 (0)	1 (4)	6 (27)	NA[d]
Long-term institution	1 (1)	0 (0)	0 (0)	1 (4)	0 (0)	NA[d]
Patients at home at least for one day, *n* (*%*)	72 (48)	11 (58)	3 (5)	27 (75)	31 (84)	< 0.001[a]
Days spent at home, *Mdn* (*range*)	0 (0 - 28)	8 (0 - 28)	0 (0 - 16)	21 (0 - 28)	24 (0 - 28)	0.001[a]
Care days with community care service[e],	***n* = 72**	***n* = 11**	***n* = 3**	***n* = 27**	***n* = 31**	
Mdn (*range*)	1 (0 - 28)	0 (0 - 24)	1 (0 - 5)	5 (0 - 28)	0 (0 - 28)	0.001[a]
Hours of care by community care service[e]	***n* = 36**	***n* = 5**	***n* = 2**	***n* = 15**	***n* = 14**	
Mdn (*range*)	34 (2 - 315)	12 (7 - 190)	6 (3 - 8)	38 (4 - 315)	23 (2 - 108)	0.111[a]
Type of community care service						
Family education/support, *n* (*%*)	35 (97)	5 (100)	2 (100)	14 (93)	14 (100)	NA[d]
Needs assessment, *n* (*%*)	31 (86)	4 (80)	1 (50)	13 (87)	13 (93)	NA[d]
Monitoring of vital signs/general condition, *n* (*%*)	29 (81)	5 (100)	2 (100)	13 (87)	9 (64)	NA[d]
Administration of medication, *n* (*%*)	25 (69)	5 (100)	0 (0)	10 (67)	10 (71)	NA[d]
Interventions related to enteral feeds, *n* (*%*)	22 (61)	5 (100)	1 (50)	13 (87)	3 (21)	NA[d]
Respiratory interventions, *n* (*%*)	18 (50)	2 (40)	0 (0)	11 (73)	5 (36)	NA[d]
Interventions related to excretion, *n* (*%*)	14 (39	1 (20)	0 (0)	6 (40)	7 (50)	NA[d]

Note. NA = Not applicable.

[a] Likelihood ratio chi-square. [b] Representing the cumulative hospital admissions in all patients. [c] Information was missing for some cases. [d] No significance testing conducted due to small numbers. [e] Consisting of nurses, most of them specialised in paediatric and/or community nursing

4.6. Discussion

There are several principal findings in this nationwide study looking at patterns of care at EOL in four distinct cohorts of diagnostic groups: patients had a variety of primary diagnoses, covering seven different ICD-10 diagnostic chapters; sixty-two percent of all patients died in ICUs, with 84% of them following a decision to withdraw life-sustaining treatment; reliance on invasive medical interventions was prevalent and patients were exposed to polypharmacy; patients experienced many symptoms with an average of six symptoms per patient; and less than half of the patients were at home at any point during their last four weeks of life with patients in the cardiology and neonatology groups having considerably fewer home days than patients in the neurology and oncology groups. However, the days of care provided by community healthcare providers were also low for these groups.

The study provides a comprehensive overview of current EOL care practices in a heterogeneous real-life setting of hospitals, long-term institutions and community healthcare organisations. The inclusion of patients with all major diagnoses responsible for disease- and prematurity-related childhood deaths and comparisons across the diagnostic groups provides additional insight and understanding for healthcare professionals. Previous studies in this field were frequently limited to the hospital setting [1, 9] or to a specific diagnostic group [10, 14, 15]. Our study is limited by its cross-sectional, primarily descriptive design incorporating a retrospective chart review. This approach does not allow conclusions to be drawn about care quality and quality of life at the EOL. Future analysis of other data from the PELICAN study will overcome some of these limitations by linking results about the parental perspective, which are reported elsewhere [17]. Known reliability issues related to chart reviews were kept to a minimum by taking established and appropriate measures. However, mixed quality of documentation among healthcare personnel resulting in incomplete or missing data still limits the data's reliability [18].

The comparisons between the four major diagnostic groups highlight elements that warrant discussion. Medication counts in our study were high, with an overall daily median of 12 drugs daily or as-needed medication orders during the last week of life. This number is higher than the reported average of 9 drugs in a study involving 515 paediatric patients with a similar diagnostic profile receiving and PPC [9], and considerably higher than the total number of 3.5 different drug categories reported in a study of 275 children during PC with diagnoses matching our neurology group [14]. We found that the medication count increased from the second-to-last week to the last week of life. Thus it seems that the intensity of medical treatment increases as the child nears death, a phase which is accompanied by a greater need for pharmacological interventions, especially for relieving pain. The high number of medications in the cardiology group is often due to frequent need for CPR and a high prevalence of surgical interventions, which are also described in other studies with cardiological patients [15, 20]. Although not perfectly comparable, symptom type and prevalence differed from the aforementioned study of 515 patients receiving PPC, in which pain was only the sixth most frequent symptom as extracted from patients' charts [9]. However, pain has been reported as the most frequent symptom in other studies with various paediatric cohorts in PC or EOL care [10, 14]. Our study adds to existing knowledge by demonstrating that symptom prevalence is dependent on the underlying CCC and that it can differ considerably.

Slightly less than 50% of our study's patients were at home at some point during their last four weeks of life. Naturally, this was only true for very few neonates. In light of the probably growing rate of prenatal diagnosis of a life-limiting CCC, early initiation of PPC may allow better planning and implementation of specialised care services at home [22]. A recent study from the US showed that 54% of hospital-based PPC programmes participating in a 2012 survey provided prenatal consultation [13].Of our study patients who spent at least one day at home, only half received community-based healthcare services. Recent data from Germany and the US show that the coordination and provision of specialised palliative home care is able to alleviate caregivers' distress and burden [23]. and to improve both the child's [24]. and the caregivers' quality of life [23]. As reported by our study, community nursing care encompasses a range of service types. The high level of coordination with the leading team in the hospital and the expertise required makes it especially challenging. Further subgroup analysis of our study's at-home population is ongoing and will target facilitators for and barriers to EOL care in the home setting.

PPC is growing internationally and the provision of consultation by a hospital-based multiprofessional PPC team seems to be the favoured model of care [13]. This requires a high level of expertise and efficient collaboration across a variety of paediatric subspecialties. The capacity of community healthcare services needs to be expanded with an emphasis on efficient coordination and continuity of care. As already recommended by an EAPC White Paper [25], outcome measurement has to be introduced into practice to evaluate quality of care and further advance research in the field.

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4.9. Declaration of conflicting interests

The Authors declare that there is no conflict of interest.

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Chapter 5

Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu) – An instrument to assess parental experiences and needs during their child's end-of-life care

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5.1. Abstract

Aim. To develop and test the Parental PELICAN Questionnaire, an instrument to retrospectively assess parental experiences and needs during their child's end-of-life care.
Background. To offer appropriate care for dying children, healthcare professionals need to understand the illness experience from the family perspective. A questionnaire specific to the end-of-life experiences and needs of parents losing a child is needed to evaluate the perceived quality of paediatric end-of-life care.
Design. This is an instrument development study applying mixed methods based on recommendations for questionnaire design and validation.
Method. The Parental PELICAN Questionnaire was developed in four phases between August 2012–March 2014: phase 1: item generation; phase 2: validity testing; phase 3: translation; phase 4: pilot testing. Psychometric properties were assessed after applying the Parental PELICAN Questionnaire in a sample of 224 bereaved parents in April 2014. Validity testing covered the evidence based on tests of content, internal structure and relations to other variables.
Results. The Parental PELICAN Questionnaire consists of approximately 90 items in four slightly different versions accounting for particularities of the four diagnostic groups. The questionnaire's items were structured according to six quality domains described in the literature. Evidence of initial validity and reliability could be demonstrated with the involvement of healthcare professionals and bereaved parents.
Conclusion. The Parental PELICAN Questionnaire holds promise as a measure to assess parental experiences and needs and is applicable to a broad range of paediatric specialties and settings. Future validation is needed to evaluate its suitability in different cultures.

5.2. Summary statements

Why is this research needed?
- Paediatric end-of-life care is challenging and complex, covers several paediatric specialties and different age groups, and has specificities other than in adult end-of-life care.
- To offer appropriate care for dying children and adolescents, healthcare professionals need to understand the illness experience from the parental perspective and to anticipate their needs.
- No valid instrument exists that allows a standardised assessment of parental experiences and needs in order to evaluate the quality of paediatric and neonatal end-of-life care.

What are the key findings?
- The Parental PELICAN Questionnaire (PaPEQu), designed to assess parental experiences and needs, aims at providing an evaluation tool for the perceived quality of family-centred paediatric and neonatal end-of-life care.
- The PaPEQu was developed based on six quality domains of family-centred care identified in the literature, providing a conceptualization of themes deemed most important to parents.
- Evidence of initial validity and reliability were demonstrated with the involvement of experienced healthcare professionals and more than 200 bereaved mothers and fathers.

How should the findings be used to influence policy/practice/research/education?
- The four slightly different versions of the PaPEQu make it applicable to a broad range of paediatric specialties and settings.
- The instrument should be further tested in populations with different cultural backgrounds and with a bigger sample for each diagnostic group.
- Future research should focus on empirical model development and testing, to facilitate the development, implementation and evaluation of appropriate interventions.

5.3. Introduction

Losing a child is a traumatic life event for parents, siblings and the extended family, in which highly stressed parents try to maintain hope while readying themselves for the loss of their child. When the death of a child is imminent, care focuses on preparing for an anticipated death while managing the end stage of a life-limiting medical condition. End-of-life (EOL) care of these vulnerable patients and families demands a comprehensive approach that includes the need for healthcare professionals to understand the illness experience from the perspective of the dying child and his/her family in order to offer appropriate care [1-3]. Prior studies describing the experiences of parents who lost a child show that open and honest communication, true relationships and adequate emotional support, symptom control and alleviation of suffering, continuity and coordination of care, and support after the death of the child are central issues for parents [2, 4]. Most of this evidence originates from qualitative research; only three quantitative studies were identified in two recent integrative reviews [2, 4]. Quality improvement initiatives risk meeting the values of healthcare professionals rather than those of parents, if the parental perspectives are not evaluated with valid measures [3, 5]. To assess and explore the parental perspective effectively, an approach combining quantitative and qualitative information is likely to provide the most comprehensive view of experiences and unmet needs [6].

5.3.1. Background

Family experiences and needs are influenced by the quality of care [6]. Healthcare quality is a complex concept, which is challenging to measure. The assessment of care quality requires indicators which can be used to develop a valid and reliable measure [3]. Existing evidence from qualitative research has led to the Initiative for Pediatric Palliative Care (IPPC) and the identification of six quality domains to guide quality improvement and development of healthcare services for families of children/adolescents with life-threatening illnesses [7, 8]. These domains include: “Holistic Care of the Child; Support of the Family Unit; Involvement of Child and Family in Communication, Decision Making, and Care Planning; Relief of Pain and Other Symptoms; Continuity of Care; and Grief and Bereavement Support” [7, p. 328] and should be addressed to enhance the capacity of children’s hospitals and home care organisations to provide high quality family-centred palliative and EOL care.

A literature review revealed few self-administered questionnaires designed to assess the experiences and needs of parents of dying children and adolescents. Meert et al. [9] developed the 68-item *Bereaved Parent Needs Assessment (BPNA)* instrument to measure parents’ needs and need fulfilment around the time of their child’s death in the paediatric intensive care unit (PICU). The instrument showed high reliability (Cronbach’s $\alpha > 0.92$) for the importance, fulfilment, and percent fulfilment scales. Criterion validity was partly demonstrated by correlating the BPNA with the *Inventory of Complicated Grief* and the *World Health Organization Quality of Life* questionnaires. The authors suggest using the instrument to assess parents’ needs and fulfilment of needs within and across PICUs rather than for individual parents [9]. Another questionnaire for PICU settings in the United States was developed by Meyer et al. [10] to examine the perspectives and priorities of parents for improved EOL care. No validity or reliability data were reported for this 28-item *Parental Perspectives Questionnaire*. In a Swedish population-based study involving parents who lost their child to cancer and focusing on parents’ self-assessment of whether they had worked through their grief and whether professional and social support facilitated this process, Kreicbergs et al. [11] developed a self-administered instrument with 129 questions and a total of 365 items. Part 1 of the questionnaire covered domains such as communication, symptom control and treatment, and issues around the impending death of the child. The questionnaire was developed involving bereaved parents and piloted to test for participation rate, failure to respond and logistics, but no validity data were reported.

Teno et al. [12] suggested that the most important aspect to evaluate quality of care is to assess whether healthcare providers responded to the expectations and preferences of the dying patient and their families. This can be achieved with asking very specific questions concerning the parents’ lived experiences [13]. Widger and Picot et al. [13] stated, that “one of the major barriers to evaluating end-of-life care provided to children and their families has been the lack of a comprehensive and sensitive instrument that can be used to collect meaningful information from families who have experienced the death of a child” [13p. 53]. Just recently Widger et al. [3] reported on the initial development and testing of an instrument to measure the quality of children’s EOL care. The 95-item instrument demonstrated initial evidence for test-retest reliability, internal consistency and construct validity for six subscales, and content validity for four additional domains. The instrument shows promise for the retrospective application in bereaved mothers. However, the applicability in bereaved fathers remains an open question [3].

None of the instruments available at the beginning of this study included all of the quality domains identified by the IPPC and were applicable to the heterogeneous field of paediatric EOL care, including children/adolescents of different age groups, with different underlying illnesses and in different clinical settings, and finally, none considered maternal and paternal perspectives. Gaining uniform information across these different situations will help identifying differences between subgroups and thus, add important and essential knowledge needed for healthcare professionals to provide individualized family-centred care during this difficult period of time.

5.4. The study

5.4.1. Aim

The purpose of this study was to develop and test the Parental PELICAN Questionnaire (PaPEQu), an instrument to retrospectively assess parental experiences and needs during their child's EOL care. This study is part of a larger study; the PELICAN study (Paediatric End-of-LIfe CAre Needs, 2012-2015, NCT 01983852). The overarching aim of the PELICAN study is to provide comprehensive information and understanding about current practices of EOL care in Switzerland (PELICAN I, retrospective chart review), about parental perspectives (PELICAN II, mixed methods) and the perspectives of the healthcare professionals (PELICAN III, focus groups) in order to formulate recommendations for best care during this highly vulnerable and critical phase of life. The PELICAN study is a nationwide study encompassing three Swiss language regions and including patients who died in the years 2011 and 2012 due to a cardiac, neurologic or oncologic illness or during the neonatal period and their parents, and healthcare professionals [14].

5.4.2. Ethical considerations

The question had been raised about whether research in bereaved families is reasonable. However, studies with bereaved subjects have shown response rates as high as 80% [11], which is consistent with our experience that parents who lost a child want to tell their story. Nevertheless we decided to choose an interval of at least 12 months after the child's death so that parents would have passed the immediate stages of grief. This is in concordance with Maciejewski et al. [15] who reported that all negative grief indicators peaked within six months post loss.

The PELICAN study was approved by the ethics committees in all of the Swiss cantons in which the study took place. Eligible parents were informed and invited to participate in the study by the former treating team, who also acted as gatekeeper as needed. Only a few families were excluded because it would have been inappropriate to invite them. Participation was entirely voluntary and written informed consent was obtained from all participants and for each study part independently. Due to the sensitive topic, psychological support was offered to potentially distraught parents.

5.4.3. Methodology

Questionnaire development followed recommendations by Streiner and Norman et al. [16] and Rattray and Jones et al. [17]. The development process was conducted in four phases: phase 1: item generation; phase 2: validity testing; phase 3: translation; and phase 4: pilot survey. Psychometric properties were assessed using the results of the PELICAN II study, quantitative part. Validity testing covered evidence based on test content, internal structure, and relations to other variables according the Standards for Educational and Psychological Testing [18].

5.4.4. Instrument development phase 1: Item generation

The first set of items, written in German, was generated from August-September 2012 within the study group and was based on scientific evidence and existing similar instruments found in the literature [9-11, 13], the expertise of group members within the field of paediatric PC and/or in the four paediatric sub-specialties (cardiology, neonatology, neurology and oncology). It became clear early in the development process that separate questionnaires for each of the four diagnostic groups (cardiology, neonatology, neurology, and oncology) were needed to account for age and illness trajectories differences between the four groups. However, item differences between the four questionnaires were kept to a minimum to preserve the opportunity to analyse data from the entire sample. The items were thematically structured according to the six quality domains grounded in the framework of the IPPC [7]. As proposed by Truog et al. [19], "Holistic Care of the Child" and "Support of the Family unit" were integrated and "Shared Decision Making" was disentangled from "Communication" and became a separate domain. Within each of the six domains, items were organised into scales concerning parental experiences and indexes for parental needs [20]. For experience-related items, 7-point adjectival response options with varying end-points anchors, *"never-always"*, *"not clear at all-very clear"*, *"not honest-honest",* or 5-point Likert-type response options where respondents indicated the extent to which they agreed with the statement were crafted. For needs-related items, 7-point adjectival indexes with end-point anchors *"not important at all-very important"* were used [16]. Content of the needs-related items was closely linked to the experience-related items within the same domain unless it was not appropriate, e.g. asking how important parental need was to alleviate pain in their child. Questions requiring a multiple choice or dichotomous response pattern were also integrated in the domains as appropriate. To summarise, parents were asked to indicate their overall satisfaction with each of the topics of the six domains on a 7-point scale with end-point anchors of *"not satisfied at all and totally satisfied"*, and a *"neutral"* label in the middle. Additionally they were asked to: (1) list three positive experiences and three negative experiences in an open-ended question; (2) indicate what areas of their personal life were negatively influenced by the illness and loss of their child by choosing all that apply from given response options; and (3) how they would rate their current overall quality of life (QoL) on a vertical visual analogue scale (VAS) ranging from 0 (worst possible) to 10 (maximum). The final section of the questionnaire included socio-demographic items. For the pilot test only, parents were asked to indicate, the level of burden of completing the questionnaire again on a (VAS) ranging from 0 (no burden) to 10 (maximum burden). For scoring, only experience-related scale items were summed to yield one experience score for each domain. All other items were treated as single items.

5.4.5. Instrument development phase 2: Validity testing – evidence based on test content

The validity of the questionnaires based on test content was assessed in three stages.

Stage 1: Item review, face validity and content validity index (first round)

In a first round, 26 German-speaking Swiss medical, nursing and psychology clinicians in the field of paediatric PC and/or working in one of the four paediatric sub-specialties (7, 7, 7, and 5 for the cardiology, neonatology, neurology, and oncology version respectively) reviewed the initial drafts of the questionnaires between October and December 2012. For each individual item, the experts were asked to rate its clarity (not clear (0) / clear (1)) and relevance (not relevant (1) / somewhat relevant (2) / quite relevant (3) / highly relevant (4)) in relation to parental experiences and needs related to their child's EOL care [21]. In addition, the experts were told that they could add comments for each item. To evaluate the agreement among experts concerning the clarity and relevance of single items, item content validity indexes (I-CVI) were calculated. The I-CVI was computed as the number of experts giving a rating of either 1 for clarity and either 3 or 4 for relevance, divided by the total number of experts. An item with an I-CVI of ≥ 0.78 for its relevance was considered to have excellent validity, whereas items with an I-CVI < 0.78 needed to be revised. As for the validity of the entire questionnaire, the average I-CVI across all items (S-CVI/Ave) [21] and the proportion of items with an I-CVI > 0.78 were calculated. For the S-CVI/Ave Polit et al. [21] recommends at least a value > 0.8, but ideally, better than 0.9. The same thresholds were also applied to the calculations for the clarity of the items. The written expert's comments were summarised for each item and taken into consideration when discussing potential revisions of the items by the study group.

Stage 2: Cognitive interviews

To ensure that the perspectives and issues relevant to parents who lost a child were captured by the questionnaire, individual cognitive interviews were conducted with four parents who lost a child in 2010. The goal of cognitive debriefing is to pre-test survey instruments by evaluating sources of response errors. It explicitly focuses on the cognitive processes respondents use to answer survey questions [22]. The parents in the current study were recruited purposefully from two tertiary care centres with the goal of having one representative of each of the four diagnostic groups. All interviews were conducted at the parents' home between April and May 2013. During the interviews the parents were asked to repeat the questions in their own words to assess comprehensibility, and they were asked to evaluate the relevance of items and single words. They were also asked about their ability to recall their child's EOL period and about the burden associated with answering the questions. The interviews were structured, using an interview guide with probing questions to gain a deeper understanding of the way respondents interpreted the questionnaire items. All interviews were audio recorded and analysed within the study group based on the written interview protocol which summarised interview content.

Stage 3: Item review, face validity (second round).

In a second round, a subsample of 7 (3 physicians, 4 nurses) was chosen from the 26 clinicians who participated in the first round to review the revised questionnaires again and to provide written feedback for the current items focusing on which domain the item was measuring and whether the concepts of parental experiences and needs during their child's EOL care were covered.

5.4.6. Instrument development phase 3: Translation

The four versions of the PaPEQu were then translated into French and Italian, the two other main languages in Switzerland, during June/July 2013. The translation process followed the guidelines of the International Society of Pharmacoeconomics and Outcome Research (ISPOR) taskforce for Translation and Cultural Adaptation [23]. The three steps applied included: (1) forward translation (conducted by a French or Italian bilingual layperson); (2) back translation (conducted by professional translators); (3) back translation review and harmonisation. The cognitive debriefing step was omitted for the French and Italian versions. The study team reasoned that potential comprehensibility issues could be uncovered and resolved during step 3 of the translation process and during the planned pilot survey. Additionally, the target population of the questionnaires in the French and Italian part of Switzerland was very small and the goal was to minimize parental burden associated with participation.

5.4.7. Instrument development phase 4: Pilot survey

In order to further identify items that lack clarity or may not be appropriate for bereaved parents, to test the recruitment strategy and logistics, and to assess the level of burden associated with filling out the questionnaire, a pilot survey was conducted in a sample of French, German and Italian speaking parents This survey was conducted in three Children's University Hospitals in the German part, one paediatric department of a University Hospital in the French part and the Paediatric Medical Centre in the Italian part of Switzerland. Purposefully chosen parents of patients who died in the year 2010 due to a cardiac, neurologic or oncologic illness or during the first four weeks of the child's life were invited to participate. The recruitment goal was to reach a stratified sample of 36 families (3 for each diagnostic group and each language). Eligible parents received an invitation letter together with the study documents from the hospital where their child had been treated. Following parental written informed consent, the families' demographic information was transmitted by the research coordinator from each site to the study team and the questionnaires were sent out from the study centre during September 2013. Each questionnaire was assigned a unique ID to enable follow-up of those who did not complete the questionnaire. Three weeks after sending out the questionnaires, a reminder card was sent to those parents who did not return their questionnaire. Non-responders to this reminder were dropped from the study.

5.4.8. Setting and sample of the PELICAN II study, quantitative part

The same recruitment processes were applied for the PELICAN II study. Beforehand, all Swiss children's hospitals, general hospitals with a paediatric unit, long-term institutions and paediatric community care services had committed to participate and execute the recruitment procedures. All parent couples of the population-based available sample of 307 deceased children were invited if; 1) their child died in the years 2011 or 2012 due to a cardiac, neurologic or oncologic illness or during the neonatal period (from the first day up to four weeks of life); 2) they could read French, German or Italian. Parents were excluded if their child died within the first 24 hours after birth. Recruitment took place in 17 hospitals, 2 long-term institutions, 4 paediatric community care services, and at 2 paediatric practitioners' practices between July 2013 and March 2014. Other than in the pilot survey, parents who did not provide written informed consent within three weeks after receipt of the study documents were called by phone, and verbal study information and clarification of potential questions was offered by the local study coordinator. Of the 307 families invited, 135 (44%) consented for the questionnaire survey (PELICAN II, quantitative part). In April 2014, questionnaires were sent out to 224 mothers and fathers who individually consented.

5.4.9. Data Analysis

For each item the percentage of missing responses was calculated. Possible reasons identified for not responding to an item were that respondent found the item objectionable or found the item too difficult to understand [16, 17]. The distribution of responses was checked by measures of central tendency and dispersion. Evidence based on internal structure was assessed through inter-item correlation, Cronbach's alpha, and exploratory factor analysis (EFA). Items within a scale should be positively and moderately correlated, after negatively formulated items are recoded. Cronbach's alpha gives the average of all possible split-half reliabilities of a scale and values above 0.7 are considered "good" for scales with fewer than seven items [16]. EFA, using principle axis factoring was applied to test for unidimensionality of the six different experience scales. Sampling adequacy for analysis was verified following the Kaiser-Meyer-Olkin measure of sampling adequacy (KMO) for individual and for multiple items. Values > 0.5 are considered mediocre, > 0.7 good and > 0.8 great. Factors were extracted based on eigenvalues > 1. Since only one factor for each scale was expected, no rotation was needed for interpretation [24]. Only scale items that were applicable to all respondents were included in reliability testing and used to calculate a scale mean. For two scales, the responses to two items that were worded differently for the neonatology and cardiology groups but were equivalent content-wise with items from versions administered to other two groups were aggregated. The scale mean was then used to calculate the Spearman's rho correlation coefficient between the corresponding single item measuring overall satisfaction of care of each of the six domains. We hypothesised that high correlations may indicate evidence on relation to other variables, i.e. criterion validity. Questionnaire responses were entered into and analysed using IBM© SPSS© Statistics 21 (IBM Corp., Armonk, NY, USA).

5.5. Results

5.5.1. Phase 1, 2 and 3: Item generation, item review and translation

The first draft of the four questionnaires for cardiology (C), neonatology (Neo), neurology (N) and oncology (O), consisted of 131/139/146/143 items respectively representing the six domains, followed by either 12 summary items for C, O and N or 10 for Neo, and 17 socio-demographic items for all versions. This added up to a total initial item count of 160/166/175/172 for the C/Neo/N/O questionnaires respectively.

Phase 2, Stage 1: Item review, face validity and content validity index (first round)

The average content validity index was > 0.78 and therefore considered excellent for all four questionnaire versions. However, when looking at single items, there were still a substantial percentage of items below the threshold of 0.78 for clarity and fewer for relevance (Table 1). Based on these results and the written feedback from the expert panel, the total number of items was reduced by approximately 50, resulting in total item count for the C/Neo/N/O versions of 109/114/117/116 respectively (including 13 socio-demographic items in all versions). Additionally, a number of items were reformulated or put in a different order.

Table 1 Average and percentage of expert agreement of the first set of items

	Clarity		Relevance	
Questionnaire	S-CVI/Ave	% of items with I-CVI < 0.78	S-CVI/Ave	% of items with I-CVI < 0.78
Cardiology	0.82	29	0.85	23
Neonatology	0.82	27	0.90	14
Neurology	0.84	24	0.87	21
Oncology	0.89	22	0.94	13

S-CVI/Ave, scale-level content validity index, averaging method; I-CVI, item-level content validity index

Phase 2, Stage 2: Summary of cognitive testing

The input from the cognitive debriefings of the four participants led to a further item reduction of 7 items (C), 11 items (Neo), 11 items (N) and 10 items (O) resulting in a second draft of the questionnaire containing 102/103/106/106 items for C/Neo/N/O respectively. The main reasons for item deletion were: that they were not relevant, not comprehensible, or too similar to other questions. The ability to recall the events around the last days/weeks of their child was not a problem for any of the participating mothers. Although remembering this time was surely painful, all of the participants stated that the burden of answering the questions was tolerable and justified by the importance of the study.

Phase 2, Stage 3 and Phase 3: Item review, face validity (second round) and translation

All experts agreed that the six quality domains structuring the questionnaires were well represented by the existing items and that the items were well formulated.

The translation process led to several small adaptations of the wording in all languages.

5.5.2. Phase 4: Pilot survey

For the pilot survey, 31 individual questionnaires (mother and father versions) were sent to 20 families covering all four diagnostic groups and all three languages. Recruitment for this pilot test taught us that the target population of parents who lost a child is limited in the Italian speaking part of Switzerland and the single recruited parent did not return the questionnaire. The receipt of 24 completed questionnaires represented a response rate of 77% and covered all diagnostic groups and the German and French languages.

The number of items, rationale for revision and examples of items dropped based on the pilot survey are presented in Table 2. The median level of burden related to completing the questionnaire, as reported by 23 parents, was 5.5 (interquartile range, 0.8-6.6) on a scale between 0 and 10. Based on this last development step, the final number of items for the C/Neo/N/O questionnaires was 91/92/95/95 respectively. A graphical illustration of the complete development process is displayed in Figure 1. A complete list of all final items (socio-demographic items excluded) for all four questionnaire versions can be accessed via http://nursing.unibas.ch/PELICAN-PaPEQU or www.kispi.uzh.ch/fzk/PaPEQu (see end of chapter). The questionnaires were then used for the main PELICAN II study.

Of the 224 questionnaires sent out, 200 were completed and sent back, representing a response rate of 89%. The frequencies according to diagnostic groups, language and mothers/fathers are presented in Table 3. The neonatology group was the largest one with 81 (41%) questionnaires followed by 48 (24%) neurology, 45 (22%) oncology and 26 (13%) cardiology questionnaires. For the total of 96 items (socio-demographic items excluded) across all four versions, the average percentage of true missing responses was at 1.8%. There were some items for which the response option “not applicable/don’t know” was present and for those items the percentage of information not available increased up to 38%. Therefore, two items were dropped from analysis completely. As a result, the scale items assessing experiences in the communication with physicians were analysed as single item as only two items remained.

5.5.3. Evidence based on internal structure and relations to other variables

Psychometric information for each scale of the six quality domains assessing parental experiences is shown in Table 4. The KMO measure verified the sampling adequacy for proceeding with EFA, only 2 of the 24 items remained under the desired threshold of 0.7. As expected, the factor solution demonstrated one factor only for each scale, supporting evidence of unidimensionality within each domain. Scale means were high in each domain and the distribution was skewed showing a ceiling effect. This was also true for the overall satisfaction ratings for each domain. All correlations between the scale mean and the corresponding satisfaction score were statistically significant with Spearman rho coefficients between 0.37 and 0.63. All other questionnaire items were analysed as single items and were not subject to reliability testing [20]. The results of the PELICAN II study, applying the PaPEQu, will be covered in future reports.

Table 2 Criteria applied to revise the PaPEQu (based on the pilot survey)

Criteria	Description	**Questionnaire domain:** Revision	Examples of removed items
Homogeneity of subscale items	As measured by inter-item correlation and Cronbach's alpha. Very high and negative inter-item correlations were undesirable and those items were removed.	**Support of the family unit:** Three items removed	*I knew to whom to turn to when in need of help.*
		Communication: Two items removed	*I understood the information about my child's condition.*
		Relief of pain and other symptoms: Two items removed	*That my child received fluids until the end. (Neo only)*
		Continuity and coordination of care: Two items removed	*My child was always admitted to the same ward for repeated hospitalisations. (C, N, O only)*
		Bereavement support: Six items removed	*My child's death has completely caught me off guard.*
Equivalence	We strove for equivalent item count and content in all four questionnaire versions as much as justifiable.	**Support of the family unit:** Two items removed, one item added	*My expertise in caring for my child was respected. (N, O only)*
		Relief of pain and other symptoms: One item removed, one item reformulated	*I needed that my child received medication to calm her/him. (C, N, O only)*
Clarity	When it was suspected that missing responses were caused by lacking clarity.	**Shared decision making:** Two items reformulated	
		Relief of pain and other symptoms: One item removed	*I needed to be able to nurse my child. (Neo only)*

C, cardiology; Neo, neonatology; N, neurology; O, oncology

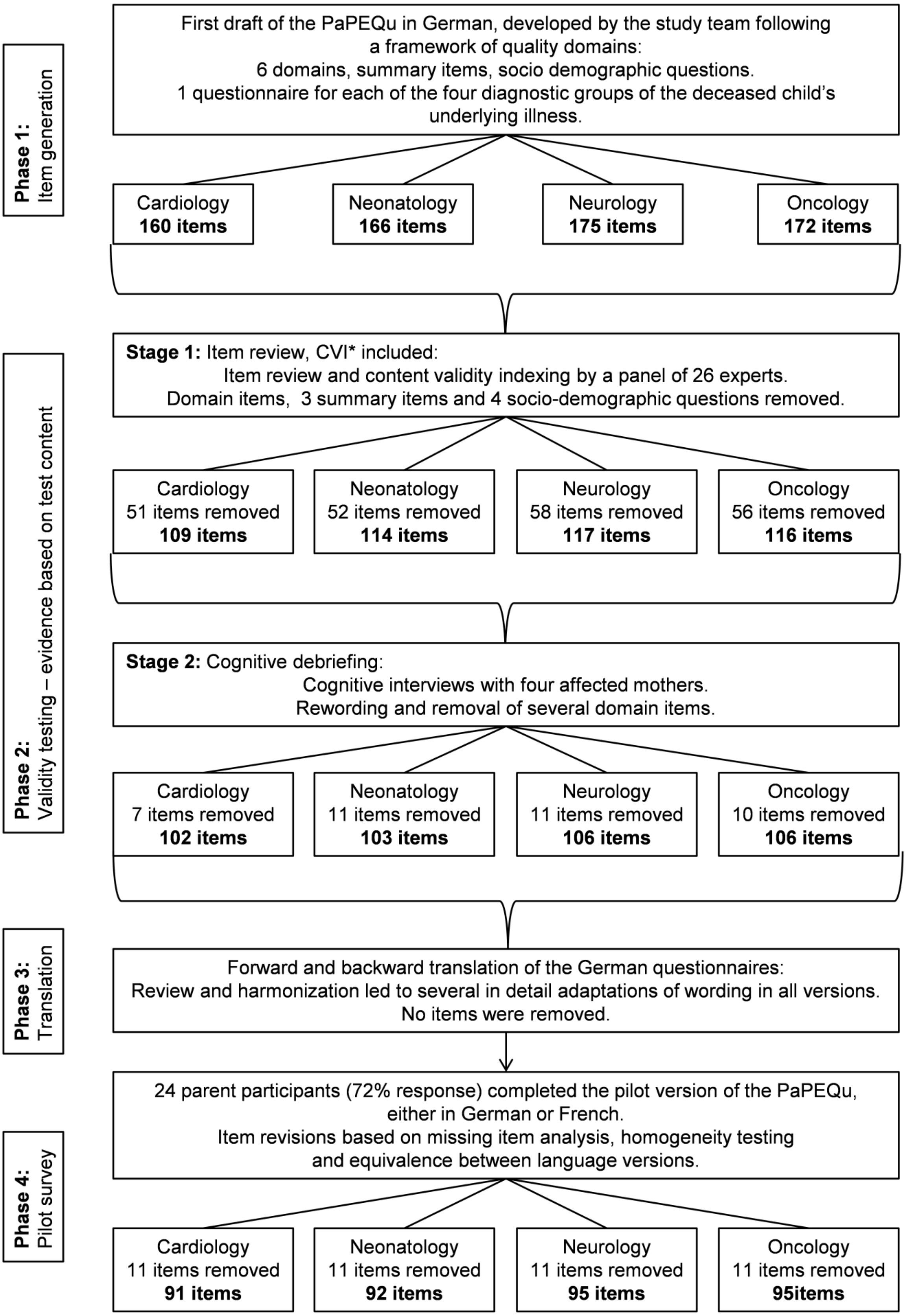

Figure 1 Detailed summery of the development of the PaPEQu.
*CVI, Content Validity Index

Table 3 Sample characteristics of the PELICAN II study, quantitative part

	Diagnostic groups				
	Cardiology n (%)	Neonatology n (%)	Neurology n (%)	Oncology n (%)	Total n (%)
Language					
German	21 (81)	66 (82)	44 (92)	31 (69)	162 (81)
French	5 (19)	9 (11)	3 (6)	12 (27)	27 (15)
Italian	0 (0)	6 (7)	1 (2)	2 (4)	9 (4)
Total	26 (100)	81 (100)	48 (100)	45 (100)	200 (100)
Parent					
Mother	14 (54)	43 (53)	28 (58)	27 (60)	112 (56)
Father	12 (46)	38 (47)	20 (42)	18 (40)	88 (44)
Total	26 (100)	81 (100)	48 (100)	45 (100)	200 (100)

Table 4 Parental experiences scale characteristics

Questionnaire domain Scoring	Number of items	Scale Mean (SD)	Cronbach's alpha	Minimum/Maximum Inter-item correlations	Kaiser-Meyer-Olkin MSA⁺	Number of factors* % of explained variance	Unrotated Factor loadings
Support of the family unit Scale range 0 - 6	4	4.97 (1.01)	0.77	0.25 – 0.70	0.75	1 61	0.40 – 0.85
Communication in general: Scale range 0 - 6	6	4.68 (1.06)	0.83	0.28 – 0.67	0.85	1 57	0.51 – 0.80
Shared decision making: Scale range 0-6	3	4.82 (1.23)	0.79	0.53 – 0.63	0.70	1 71	0.68 – 0.81
Relief of pain and other symptoms: Scale range 0-6	3	4.99 (1.10)	0.88	0.62 – 0.78	0.71	1 81	0.76 – 0.95
Continuity and coordination of care: Scale range 1-5	4	3.86 (0.91)	0.69	0.31 – 0.46	0.72	1 53	0.57 – 0.64
Bereavement support: Scale range 1-5	4	4.30 (0.79)	0.75	0.33 – 0.62	0.71	1 58	0.54 – 0.86

⁺ Measure of Sampling Adequacy: Values for the 24 single items ranged between 0.65 and 0.89; * Based on exploratory factor analysis and eigenvalues > 1.

5.6. Discussion

In this article we describe the development and validation of a questionnaire designed to retrospectively assess parental experiences and needs during EOL care of their child. The process followed existing recommendations and applied various rigorous quantitative and qualitative methods. The PaPEQu aims at providing an evaluation tool of perceived quality of family-centred paediatric EOL care. Evidence of initial validity and reliability were demonstrated using a sample of experienced healthcare professionals and more than 200 bereaved mothers and fathers. The four slightly different versions of the PaPEQu make it applicable to a broad range of paediatric specialties and settings. The questionnaire's items were structured according to six quality domains described in the literature, providing a conceptualisation of themes deemed most important to parents during the EOL phase of their child. In a recently developed and tested instrument to measure the quality of children's EOL care, 10 domains arose from EFA [3]. While the domains were labelled differently, most content was actually overlapping which strengthens the validity of the items chosen for the PaPEQu. Within the six PaPEQu domains, some questions assess experiences, while others assess needs related to the experiences of parents. Parental experiences reflect the quality of care provided and might be linked to parent satisfaction. However, assessing parental satisfaction with standard satisfaction scales may not be appropriate as parents may have a low level of expectation and are easily satisfied [13]. Although all the experience scores from the six domains were significantly correlated with the corresponding single item measuring overall satisfaction of care, interpretation has to be done cautiously. The ratings of most adjectival and Likert-type items were high in our study sample and considerable ceiling effects were present. Little variability existed in the scores between the domains to render those correlations meaningful. Nevertheless, on the single item level, the full range of possible response options was used and the required variation for psychometric testing was present. EFA and Cronbach's alpha demonstrated the unidimensionality and internal consistency of the experience scale within each domain, indicating that these scale items represent a single construct consistently. However, to test how well the items represent the six quality domains would need confirmatory factor analysis and thus requiring a bigger study sample.

A questionnaire response rate of 89% was achieved within the PELICAN II study, quantitative part after 49% of the invited parents agreed to participate in the study. High response rates have been achieved in other studies with bereaved family members [11]. This is however, not always the case and the added burden that research participation imposes during bereavement should not be underestimated. Low response rates, e.g. 22%, 18.6%, have been reported elsewhere [3, 9]. Our high response rate together with the low percentages of missing items indicates that the items are clear and appropriate, considering that the burden for our participants was far from zero with a median of 5.5 on a scale from 0 to 10.

One limitation of the study is related to the sample diversity concerning culture and ethnicity. As part of the inclusion criteria participants had to be proficient in reading one of the three main Swiss languages. This proficiency is often not present in Switzerland's migrant residents (25%) resulting in the exclusion of a group of bereaved parents, which mostly represented ethnic minorities. The questionnaire's usefulness in this population therefore remains unclear and should be specifically tested before application.

5.7. Conclusions

Developing, testing and refining a new instrument is an iterative process and requires a series of studies. Although the PaPEQu will need some revisions for future use, the instrument has promise as a method to assess parental experiences and needs during their child's end-of-life. Information about specific needs is required to plan and implement needs-driven specialized care models. Quality improvement initiatives for paediatric palliative care are happening in many countries and the body of research within this field is growing. Evaluating the quality of care, requires a measure that is grounded in a sound conceptual model, which captures the reality of affected families and healthcare professionals, and explains the mechanisms of structures, processes and outcomes. Future research should focus on empirical model development and testing, in order to being able to develop, implement and evaluate appropriate interventions.

5.8. Acknowledgments

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5.10. Conflict of interest

No conflict of interest has been declared by the authors.

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List of items of all PaPEQu versions*

Support of the family unit domain	**Presence in questionnaire version**			
Items assessing parental experience: *7 point adjectival scale, never – always*	**Card**	**Neo**	**Neur**	**Onc**
(1) I felt taken seriously with the difficult situation my child and I were in.	✓	✓	✓	✓
(2) I could be involved in my child’s care as much as I wanted to be.	✓	✓	✓	✓
(3) a. When I needed respite there was a professional (e.g. nurse, volunteer) who could take my place.	✓	✓	✓	✓
(3) b. My privacy was respected.	✓	✓	✓	✓
(4) The care team was aware of my worries and fears.	✓	✓	✓	✓
(5) I trusted the healthcare professionals.	✓	✓	✓	✓
(6) What services were offered to you or your child during the child’s last four weeks of life? *List of 10 possible services to choose all those applicable + Other.*	✓	✓	✓	✓
Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
I needed				
(7) a. To have a place to sleep in the hospital close to my child. Response option *“not applicable”* available	✓	✓		
(7) b. To be involved in my child’s care.			✓	✓
(8) a. To have a room where my family and I could spend some private time together.	✓	✓		
(8) b. To have respite from the care of my child.			✓	✓
(9) To share my fears and worries with someone from the healthcare team.	✓	✓	✓	✓

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

	Communication domain	Presence in questionnaire version			
	Items assessing parental experiences: *7 point adjectival scale, never – always*	**Card**	**Neo**	**Neur**	**Onc**
(10)	Information concerning my child's condition was provided appropriately.	✓	✓	✓	✓
(11)	Bad news was communicated in a sensitive manner.	✓	✓	✓	✓
(12)	Information I received was contradictory.	✓	✓	✓	✓
(13)	My questions were taken seriously by the healthcare team.	✓	✓	✓	✓
(14)	To get information concerning my child's condition I had to ask.	✓	✓	✓	✓
(15)	I had the impression that the information I received about treatment options was complete.	✓	✓	✓	✓
	How did you experience communication with the attending physicians? *7 point adjectival scale, not clear at all – very clear*				
(16)	In general: Additionally: *7 point adjectival scale, not honest – honest*	✓	✓	✓	✓
(17)	About treatment options to alleviate symptoms:	✓	✓	✓	✓
(18)	About the prospects and limitations of life-sustaining measures: Additionally: *7 point adjectival scale, not honest – honest* Response option *"not applicable"* available	✓	✓	✓	✓
(19)	About what kind of physical changes to expect when my child is dying: Additionally: *7 point adjectival scale, not honest – honest* Response option *"not applicable"* available	✓	✓	✓	✓
(20)	Were you informed that your child could die? *Yes – No*	✓	✓	✓	✓
(21)	a. When were you informed that your child could die? Multiple choice: *A few days before my child died – Less than 4 weeks before my child died – Less than 6 months before my child died – More than 6 months before my child died*			✓	✓
(21)	b. When were you informed that your child could die? Multiple choice: *Before the birth of my child – Less than 24 hours before my child died – Less than 1 week before my child died – 2 to 4 weeks before my child died*		✓		
(21)	c. When were you informed that your child could die? Multiple choice: *Before the birth of my child – Less than 24 hours before my child died – A few days before my child died – Less than 4 weeks before my child died – Less than 6 months before my child died – More than 6 months before my child died*	✓			
(22)	Who informed your child that she/he could die? Multiple choice: *It was not possible to inform my child – Myself or the other parent – A physician together with me or the other parent – A physician alone – My child did not want to speak about it – I did not want anyone to speak to my child about dying.*			✓	✓

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

	Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
	I needed				
(23)	To have the opportunity to ask questions at all times.	✓	✓	✓	✓
(24)	To be continuously informed about my child's condition.	✓	✓	✓	✓
(25)	To find out how my child would die.	✓	✓	✓	✓
(26)	a. To be informed early about my child's imminent death.	✓	✓		
(26)	b. To be supported in maintaining hope despite the hopeless situation.			✓	✓
	Shared decision making domain	**Presence in questionnaire version**			
	Items assessing parental experiences: *7 point adjectival scale, never – always*	**Card**	**Neo**	**Neur**	**Onc**
(27)	I was involved in taking decisions.	✓	✓	✓	✓
(28)	I was sufficiently informed to contribute to the decisions taken.	✓	✓	✓	✓
(29)	I had the opportunity to question prior decisions and to re-discuss them.	✓	✓	✓	✓
(30)	Was a decision taken concerning the potential need to resuscitate your child? *Yes – No*	✓	✓	✓	✓
(31)	Who decided in favor or against potential resuscitation of your child? Multiple choice: *The topic was never discussed – Me – The other parent – Us as family – The healthcare team – My family together with the healthcare team – Someone else*	✓	✓	✓	✓
(32)	a. Was the cessation of non-helpful treatments discussed with you? *Yes – No*			✓	✓
(32)	b. Was the cessation of life-sustaining measures discussed with you? Yes – No		✓		
(33)	a. Who decided in favour of or against the cessation of non-helpful treatments? Multiple choice: *The topic was never discussed – Me – The other parent – Us as family – The healthcare team – My family together with the healthcare team – Someone else*			✓	✓
(33)	b. Who decided in favour of or against the cessation of life-sustaining measures? Multiple choice: *The topic was never discussed – Me – The other parent – Us as family – The healthcare team – My family together with the healthcare team – Someone else*		✓		
(34)	Did you receive written documentation concerning these decisions? *Yes – No*			✓	✓

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

	Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
I needed					
(35)	To be involved in taking decisions.	✓	✓	✓	✓
(36)	That my personal beliefs and values were considered when taking decisions.	✓	✓	✓	✓
(37)	Not to have the feeling that I had to take decisions all by myself.	✓	✓	✓	✓
(38)	a. That the cessation of life-sustaining measures was discussed with me.		✓		
(38)	b. That the measures to resuscitate my child were discussed with me.	✓			
(38)	c. That the cessation of non-helpful treatments was discussed with me			✓	✓

Relief of pain and other symptoms domain		**Presence in questionnaire version**			
	Items assessing parental experiences: *7 point adjectival scale, never – always*	**Card**	**Neo**	**Neur**	**Onc**
(39)	It was my impression that my child's discomforts were eased in the best way possible.	✓	✓	✓	✓
(40)	My child's pain was recognized.	✓	✓	✓	✓
(41)	My child's pain was adequately treated.	✓	✓	✓	✓
(42)	Which of the following 3 of your child's discomforts stressed you the most? List of 12 to 17 different symptoms of discomfort to choose from and rate in descending order.	✓	✓	✓	✓
	Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
I needed					
(43)	That my child received enough medication to ease her/his suffering.	✓		✓	✓
(44)	To have physical contact with my child.		✓		
(45)	That my child was awake and receptive enough to be able to play/speak/or do things with us or other people around.			✓	✓
(46)	That my child received medication to calm her/him.		✓		
(47)	That my child received complementary and alternative medicine.	✓		✓	✓
(48)	To be able to use non-pharmacological measures to ease my child's suffering, e.g. massage, tucking.		✓		
(49)	To take my child in my arms.	✓			
(50)	That my child received fluids until the end.	✓		✓	✓
(51)	That I could give my child milk, either through the tube, with a bottle or a cotton swab.		✓		

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

	Continuity and coordination of care domain	**Presence in questionnaire version**			
	Items assessing parental experiences: *5 point Likert scale, strongly disagree - rather disagree – agree partly – rather agree – strongly agree* Response option *“not applicable”* available	**Card**	**Neo**	**Neur**	**Onc**
(52)	There was a healthcare professional who coordinated my child’s care.	✓	✓	✓	✓
(53)	In the hospital, there was a physician in charge of our case whom I could always talk to.	✓	✓	✓	✓
(54)	a. In the hospital, there was a nurse in charge of our case whom I could always talk to.	✓	✓		
(54)	b. At home, my child’s care was mostly provided by the same nurse.			✓	✓
(55)	Information concerning the condition of my child was appropriately shared among the whole healthcare team.	✓	✓	✓	✓
(56)	Who mainly supported you in the organization of your child’s care during her/his last four weeks of life? Multiple choice: *List of 10 possible professionals and combinations to choose the most applicable*	✓		✓	✓
	Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
	I needed				
(57)	To have a professional from the healthcare team to coordinate the care of my child.	✓	✓	✓	✓
(58)	To have the same physician providing care.	✓	✓	✓	✓
(59)	That my child’s care was mostly provided by the same nurses.	✓	✓	✓	✓

	Bereavement support domain	**Presence in questionnaire version**			
	Items assessing parental experiences: *5 point Likert scale, strongly disagree - rather disagree – agree partly – rather agree – strongly agree* Response option *“don’t know”* available	**Card**	**Neo**	**Neur**	**Onc**
(60)	I could spend as much time with my dying child as I wanted to.	✓	✓	✓	✓
(61)	a. I received the necessary support from the healthcare team so that my child could die where I wished.	✓		✓	✓
(61)	b. I was supported by the healthcare team in creating mementos of my child.		✓		
(62)	I could say goodbye to my child in the way I wanted to.	✓	✓	✓	✓
(63)	There was solace around my child’s death	✓	✓	✓	✓
(64)	a. Where did your child pass away? Multiple choice: *In the paediatric intensive care unit – In the adult intensive care unit – On a ward in the paediatric hospital – On a ward in an adult hospital – At home – At another place*	✓		✓	✓
(64)	b. Where did your child pass away? Multiple choice: *In the neonatal intensive care unit - In the paediatric intensive care unit - On a ward in the paediatric hospital - At home – At another place*		✓		
(65)	Were you with your child when she/he passed away? *Yes - No*	✓	✓	✓	✓

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

(66)	Were siblings, other family members and/or friends with your child when she/he passed away? *Yes - No*	✓	✓	✓	✓
(67)	Did you wash and dress your child or help others to do it after she/he passed away? *Yes - No*	✓	✓	✓	✓
(68)	Were you in contact with someone from the healthcare team during the first weeks after the death of your child? *Yes - No*	✓	✓	✓	✓
(69)	Has there been a follow-up meeting with someone from the healthcare team? *Yes - No*	✓	✓	✓	✓
(70)	Could you please tell us what kind of support services you used or still use during your bereavement? *Free text field to describe the kind of service(s).*	✓	✓	✓	✓
	Items assessing parental needs: *7 point adjectival scale, not important at all – very important*				
I needed					
(71)	To have the choice of where child might die. Response option *"not applicable"* available	✓	✓	✓	✓
(72)	That family and friends could say goodbye to my child.	✓	✓	✓	✓
(73)	That I was supported by the healthcare team to structure the hours after the death of my child according my needs.	✓	✓	✓	✓
(74)	To take my child home after her/his death so that family and friends could say goodbye. Response option *"not applicable"* available	✓	✓	✓	✓
(75)	That someone from the healthcare team attended my child's funeral or burial. Response option *"not applicable"* available	✓	✓	✓	✓
(76)	To stay in contact with someone from the healthcare team after my child's death.	✓	✓	✓	✓
Summary		**Presence in questionnaire version**			
	Overall, how satisfied were you with the: *7 point adjectival scale, not satisfied at all – neutral - satisfied*	**Card**	**Neo**	**Neur**	**Onc**
(77)	Support for you and your family?	✓	✓	✓	✓
(78)	Communication with you and your family?	✓	✓	✓	✓
(79)	Shared decision-making?	✓	✓	✓	✓
(80)	Relief of pain and other symptoms?	✓	✓	✓	✓
(81)	Continuation and coordination of care?	✓	✓	✓	✓
(82)	Bereavement support	✓	✓	✓	✓
(83)	Could you please list three positive experiences concerning the care you and your family received: *Free text field to list up to three examples.*	✓	✓	✓	✓
(84)	Could you please list three negative experiences concerning the care you and your family received: *Free text field to list up to three examples*	✓	✓	✓	✓
(85)	What areas of your personal life were negatively influenced by the illness and death of your child? *List of 7 potential areas to choose from; choose all those applicable + Other.*	✓	✓	✓	✓

*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

(86)	How would you rate your current quality of life on the following scale? *Vertical visual analogue scale ranging from 0 (worst possible) to 10 (maximum).*	✓	✓	✓	✓
(87)	Is there something else you would like to tell us? *Free text field to write.*	✓	✓	✓	✓

Note. The 13 socio-demographic items are excluded from this list.
*Card, cardiology; Neo, neonatology; Neur, neurology; Onc, oncology.

Chapter 6

When parents face the death of their child: A nationwide cross-sectional survey of parental perspectives on their child's end-of life care

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6.1. Abstract

Background: Parents facing the death of their child have a strong need for compassionate professional support. Care services should be based on empirical evidence, be sensitive to the needs of the families concerned, take into account the heterogeneity within the medical field of paediatrics, and fit into the local health care system. We need to better understand the perspectives of parents facing the death of their child in order to guide further development and evaluation of specialised paediatric palliative and end-of-life (EOL) care services.

Methods: Questionnaire survey to assess the EOL care perspectives of a Swiss population-based sample of bereaved parents who had lost a child due to a cardiac, neurological or oncological condition, or during the neonatal period in the years 2011 or 2012. The parental perspective was assessed with a newly developed and tested instrument that was structured according to six evidence-based quality domains. Responses regarding parental experiences and perceived satisfaction are described. Differences between the four diagnostic groups are analysed using a generalized estimation equation to account for the dyadic data structure.

Results: Of 307 eligible families, 267 could be contacted and 135 (51%) consented to participate in this questionnaire survey. Our findings show positive parental experiences of their child's EOL care and high perceived satisfaction with the care their child received. Parents of a child with cancer rated their experiences highest in most of the six quality domains and reported the highest satisfaction with care. The lowest scores were mainly reported by parents from the neurology group, with the exception of the shared decision making domain, where parents of neonates reported significantly less positive experiences.

Conclusions: Although positive in general, our study results suggest some areas for improvement. The integration of specialised paediatric palliative care has the potential to minimise lost opportunities to support and assist parents.

6.2. Background

When facing the death of their child, parents experience an unimaginably painful life event and severe crisis that affects the whole family for life. In this highly stressful time parents are confronted with uncertainty and are required to make difficult decisions, e.g. withdrawal of life-sustaining interventions. Their need for compassionate professional support is high. Support throughout terminal care and after the loss of a child was reported to have a positive impact on long-term grieving outcomes of parents who lost a child to cancer [1]. For clinicians it is therefore imperative to know how parents experience their child's end-of-life (EOL) and what their specific needs are in order to provide good quality care. Two recent integrative reviews and a qualitative metasummary extracted existing evidence from 36 studies (29 qualitative, 7 quantitative) about parental perspectives on their child's palliative care (PC) or EOL care [2-4]. This evidence provides an overview of themes/domains most important to parents and can be summarised as: sincere relationships and emotional, spiritual and cultural support; genuine communication; alleviation of suffering; continuity, coordination and accessibility of care; and bereavement support [2-4]. Deficiencies in meeting parental needs were identified across all themes, e.g. insufficient communication, lack of respect, and lack of emotional support [2].

Caring for a child at the end of her/his life and supporting the family is most challenging for health care professionals. It requires a skilled multidisciplinary health care team that adopts a comprehensive and integrative care approach [5]. This has led to the emergence of the medical subspecialty of paediatric palliative care (PPC), which is defined by the World Health Organization as the active total care of the child's body, mind and spirit, and involves giving support to the family. It requires a broad approach that includes the family. It can be provided in tertiary care facilities, in community health centres and even in children's homes [6]. Many countries have recognized the need for PPC and a series of hospital-based programs have been developed and implemented during the last decade [7, 8]. In Switzerland, this need is acknowledged by the Federal Office of Public Health by incorporating it in its national strategy and conception for implementation [9, 10]. Their proposed framework emphasizes the importance of a person-centred approach focusing on the complexity of the situation and needs of the person concerned [10]. In the field of paediatrics especially, personcenteredness must be extended to family-centeredness, with the child and family as the unit of care. Paediatric care encompasses the whole age continuum from infants and children who have never experienced or expressed preferences to adolescents able to discuss their situation and express expectations [11], and it takes place in various in- and outpatient care settings and at home.

Fortunately, childhood deaths are a rare event. In Switzerland 424 deaths in children (0 to 14 years of age) were registered in 2013. Mortality data from developed countries show, that perinatal conditions contribute to 50% of all deaths in the first year of life. Beyond the first year, external causes, e.g. accidents, are the most common causes of death. Complex chronic conditions such as genetic/congenital disorders, neurological and cardiac conditions, and cancer represent the main causes of disease-related deaths [12]. This wide variety of underlying medical conditions leads to vastly different illness trajectories and lifespans potentially influencing what parents experience during their child's EOL care. There is little evidence concerning the influence of the child's underlying diagnosis on the parental perspective. It has been suggested that different challenges arise and that families from the oncology group can generally draw on a better developed professional support infrastructure than other affected families [13, 14].

In many of the existing studies covering parental perspectives of their child's PPC or EOL care, samples were limited either by case numbers, the inclusion of underlying illnesses causing the child's death (i.e. predominantly parents of children with cancer [13]) or the care setting (e.g. paediatric intensive care unit) [15, 16]. Care services should be based on empirical evidence, sensitive to the needs of the families concerned, take into account the heterogeneity within the medical field of paediatrics, and should fit the local health care system. We need to better understand the perspectives of parents facing the death of their child in order to guide further development and evaluation of specialised PPC and EOL care services. It was therefore the purpose of this study to assess the perspectives of bereaved parents who had lost a child due to a cardiac, neurological or oncological condition, or during the neonatal period in order to (1) describe specific parental experiences in relation to the underlying medical condition causing the child's death, and (2) explore differences in parental perspectives between four common medical conditions responsible for childhood death.

6.3. Methods

6.3.1. Design, setting, participants, and recruitment

The cross-sectional questionnaire survey was embedded in a larger research project concerned with paediatric EOL care needs in Switzerland (Paediatric End-of-LIfe CAre Needs – PELICAN, 2012-2015, NCT01983852) drawing from a population based sample of deceased children, their bereaved parents and health care professionals. The PELICAN study aimed to provide comprehensive information and understanding about the current practices of EOL care (in this study, defined as the last 4 weeks of life prior to death) in paediatric settings in Switzerland (hospital and community care), and about the perspectives of the parents and health care professionals involved [17]. The questionnaire survey reported on here covered the quantitative assessment of parental perspectives by including parents of all children that had died due to a cardiac, neurological or oncological condition or during the neonatal period in the years 2011 and 2012. These four groups were chosen, as they represent the major diagnoses causing illness-related death in children [18]. Eligible parents were identified using administrative death data from all Swiss children's hospitals, general hospitals with a paediatric unit, long-term institutions and paediatric community care services. All institutions with probable events of death were informed of the study and committed to participate and execute the recruitment procedures, which involved sending out an invitation letter together with the informed consent documents. Parents were not invited if their child had died during the first 24 hours of her/his life. Parents were included if they consented to participate and were proficient in the German, French or Italian language. Once parents sent back their written consent, their demographic information was then transmitted to the research team. If written consent was not received three weeks after receipt of the study documents, the family was telephoned by a local study coordinator to provide verbal study information and to clarify potential questions. Recruitment occurred between July 2013 and March 2014 in 8 children's hospitals (5 of them tertiary paediatric care centres), 9 general hospitals with a paediatric unit, 2 long-term institutions, and 4 paediatric community care services. For two families, the hospital delegated recruitment to a paediatric practitioner's practice which then invited the family.

Human Research Ethics Committees from the 11 Swiss cantons in which the recruiting institutions were located approved the PELICAN study (leading committee: KEK ZH Nr. 2012-0537, additional file 1) [19].

6.3.2. Measures

To retrospectively assess the parental perspective on the child's EOL care, a survey instrument, the Parental PELICAN Questionnaire (PaPEQu) was developed by the PELICAN study group. A detailed description of the development and validation of the PaPEQu has been published elsewhere [19], and a complete list of items with corresponding response options is provided (additional file 2, chapter 5). Initial validity and reliability of the PaPEQu were demonstrated in a sample of health care professionals and bereaved parents [19].

Four slightly different versions for the four diagnostic groups (cardiology, neonatology, neurology, and oncology) were created to account for differences in illness trajectories between the groups. The PaPEQU is thematically structured following the framework of six quality domains identified by the Initiative for Pediatric Palliative Care [20] and adapted by Truog et al. [21]. The six domains are in accordance with existing evidence and include: *support of the family unit, communication, shared decision making, relief of pain and other symptoms, continuity of care, and bereavement support*. Within each domain, the items were organised into scales or single items related to parental experiences and indexes for parental needs. The item count of experience related items ranged from 44 to 48 items, depending on the diagnostic group version. With 34 needs-related items and 13 socio-demographic items, the total item count of the PaPEQu ranged from 91 to 95 items. In this article, we report on the items related to parental experiences and socio-demographic information only. The results of the needs-related items showed high ceiling effects and little variation across the four diagnostic groups, making a thorough interpretation of these results difficult. Those items can still be checked in the complete item list (additional file 2, chapter 5).

For experience-related scale items, the response option was either a 7-point (0 to 6) with varying end-point anchors (*"never-always"*, *"not clear at all-very clear"*, *"not honest-honest")*, or a 5-point Likert-type (1 to 5), where respondents indicated the extent to which they agreed with the statement. The assessment of parental experiences was supplemented with single items with multiple choice or dichotomous response options (Yes-No) as appropriate. In addition, parents were asked to rate their perceived overall satisfaction with the care their child received for each of the six quality domains on a 7-point scale (1 to 7) with end-point anchors *"not satisfied at all -satisfied"* and a *"neutral"* label in the middle. They were also asked to list three positive and three negative experiences associated with their child's EOL care and to indicate what areas of their lives were negatively influenced by their child's illness and death with a question allowing multiple responses. Finally, on a 0 (worst possible) to 10 (best possible) vertical visual analogue scale they were asked to rate their current quality of life (QoL). Socio-demographic information was collected at the end of the questionnaire. Scale items were summed and averaged to yield one score per domain with higher values representing more positive experiences. The unidimensionality of the parental experiences score for each domain separately was demonstrated with exploratory factor analysis; internal consistency testing showed Cronbach's alpha levels between 0.69 – 0.88 [19].

6.3.3. Study procedures

The PaPEQu was sent out in April 2014 to mothers and fathers who individually consented to participate in this part of the PELICAN study. An identification code was assigned to each questionnaire allowing mapping of the family dyad. Parents who had not sent back the completed questionnaire within three weeks received a reminder card. Non-responders to this reminder were dropped from the study. All questionnaires were hand checked for completeness upon receipt, and electronically scanned to be downloaded onto an IBM© SPSS© data file.

6.3.4. Data analysis

The responses to all items were explored using measures of central tendency and dispersion. Descriptive statistics were used to summarize parental experiences for the total sample as well as for the four diagnostic groups. For each item the percentage and pattern of missing responses were calculated and explored using missing value analysis. Items that more than 30% of respondents either did not answer or responded to with "not applicable"/"don't know" were only analysed descriptively.

Scale items related to parental experiences that were present in all four questionnaire versions were used to calculate a scale score as described above. All other items were analysed as single items. Since most responses were skewed showing a ceiling effect, data transformation was applied. i.e. base 10 logarithm, square root, or reciprocal as indicated [22], for continuous dependent variables. Correction of severe distributional violations such as the presence of outliers was achieved. To explore differences in parental perspectives between the four diagnostic groups, various statistical tests were applied. For the experiences scores and the perceived satisfaction scores as continuous dependent variables, and for items with a dichotomous response (Yes-No), the method of generalized estimating equations (GEE) was used. GEE is an extension of the generalized linear model and allows analysing data with correlated residuals, i.e. clustered data [23, 24]. Data was clustered due to the dyadic design with correlated data between partners (mother and father), based on partner effects and common fate [25]. Diagnostic group was the model's predictor (factor with four levels) with the neonatology group set as reference for comparison since it was the group with the most cases. To control for potential gender effects independent of the dyadic structure, gender (female/male) was specified as a confounder. For items with multiple choice response options, Pearson's chi-square or Fisher's exact test were calculated as appropriate. Contribution to a potential significant main effect was interpreted by breaking down the standardized residuals with values outside ±3.29 representing significance at $P < 0.001$ [26]. To adjust for multiple testing, a probability of ≤ 0.001 was set to decide significance. All quantitative data were analysed using IBM© SPSS© Statistics 21 for Mac® (IBM Corp, Armonk, NY, USA).

Text responses from the three open-ended questions concerning support services in bereavement, and positive and negative experiences were imported in the text management program ATLAS.ti7, 7.5.4 for MS Windows® (ATLAS.ti GmbH, Berlin, Germany). These qualitative data were coded deductively, following the questionnaire's six quality domains, by two trained research assistants independently. The two solutions were merged based on consensus between the two coders. To summarise this information on the item level, frequencies of codes within the domains were counted to demonstrate which domains were prominently presented.

6.4. Results

Flow of study recruitment and participation is displayed in Figure 1. The participation rate was 51% ($n = 135$) of the 267 eligible families who received the study documents. Study participation differed between the diagnostic groups with parents from the neonatology group showing the lowest rate and parents from the oncology group the highest (Figure 1). Of the 224 individual questionnaires (mothers and fathers) sent out to the 135 families, 200 questionnaires from 124 families were completed and sent back, representing a questionnaire response rate of 89%. Parents of deceased neonates represented the largest group, followed by parents from the neurology, oncology and cardiology group (Table 1). The sample mainly consisted of Swiss residents (87%), with 13% migrant families. With neonates comprising the largest group of deceased children, most deaths occurred on a neonatal intensive care unit (ICU), as reported by the parents. The age of the deceased child differed among the diagnostic groups, with neonates obviously being the youngest (Table 1).

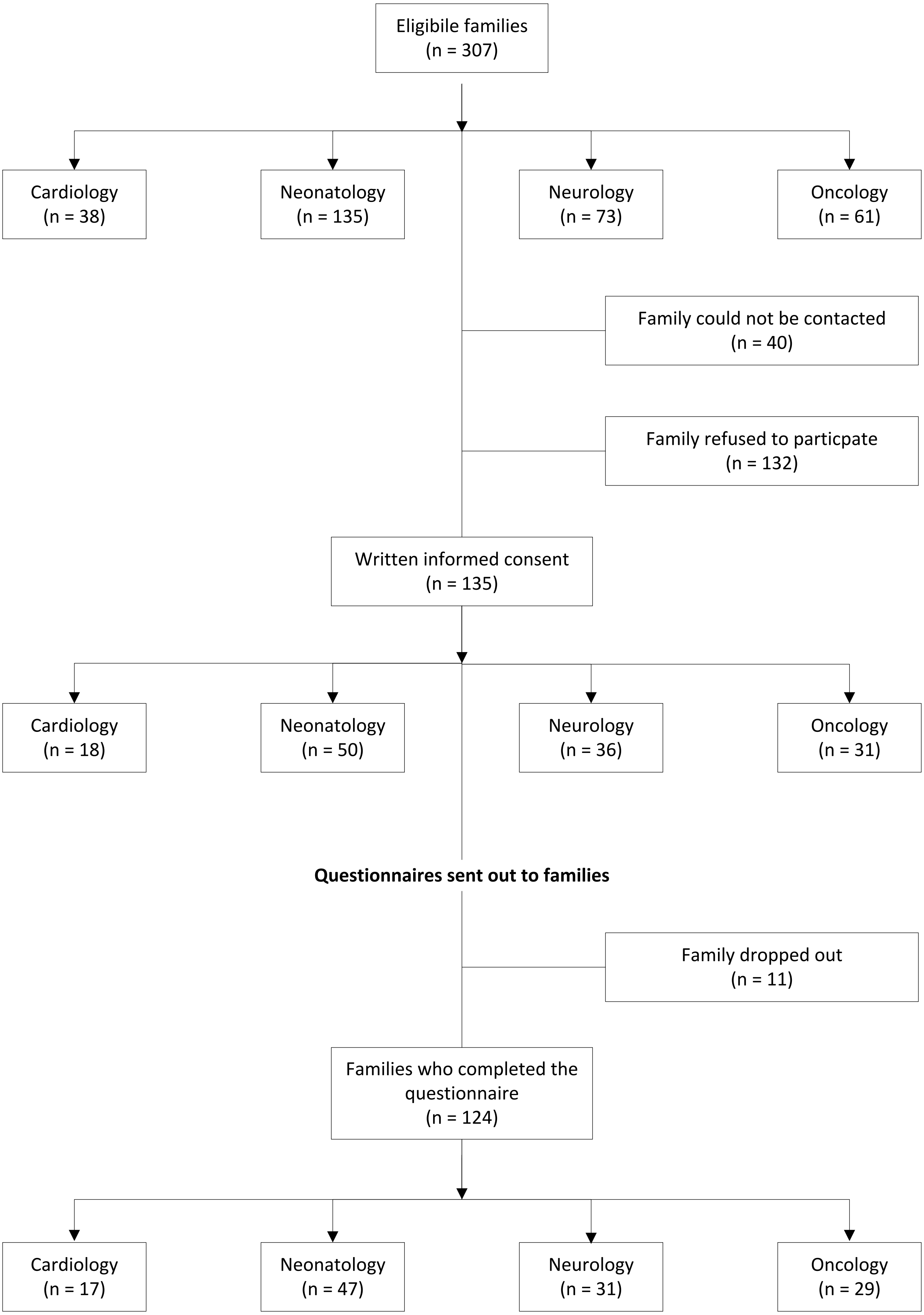


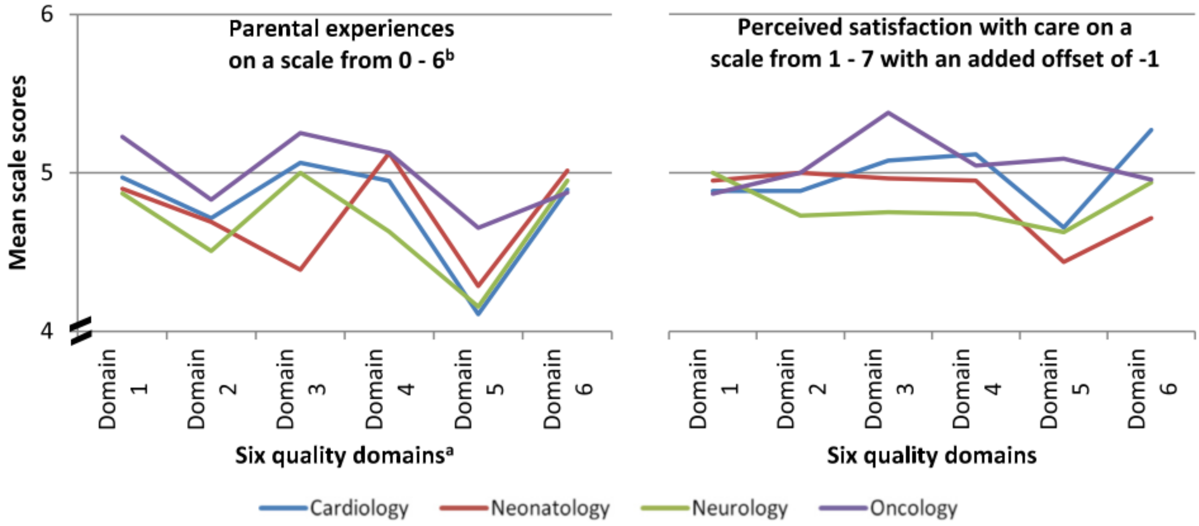
Fig 1. Study recruitment and participation

Table 1 Sample characteristics of parents (*N* = 200), place of death and age of child (*N* = 124)

Characteristics	**Cardiology** *n* = 26 (13%)	**Neonatology** *n* = 81 (41%)	**Neurology** *n* = 48 (24%)	**Oncology** *n* = 45 (22%)	**Total** *N* = 200 (100%)
Age[a], *M* (*SD*)					40 (6.48)
Mothers, *n* = 112 (56%)	38 (4.38)	37 (4.29)	41 (6.07)	43 (7.30)	39 (6.05)
Fathers, *n* = 88 (44%)	40 (6.88)	39 (5.77)	42 (6.56)	48 (5.85)	42 (6.83)
Language, *n* (%)					
German	21 (80.8)	66 (81.5)	44 (91.7)	31 (68.9)	162 (81.0)
French	5 (19.2)	9 (11.1)	3 (6.3)	12 (26.7)	29 (14.5)
Italian	0 (0.0)	6 (7.4)	1 (2.1)	2 (4.4)	9 (4.5)
Marital status, *n* (%)		n = 80			*N* = 199
Married /Partnership	22 (84.6)	79 (98.8)	43 (89.6)	41 (91.1)	185 (93.0)
Divorced/Separated	4 (15.4)	1 (1.3)	4 (8.3)	2 (4.4)	11 (5.5)
Single	0 (0.0)	0 (0.0)	1 (2.1)	2 (4.4)	3 (1.5)
Religious affiliation, *n* (%)		*n* = 80		n = 44	*n* = 198
Catholic	7 (26.9)	37 (46.3)	21 (43.8)	14 (31.8)	79 (39.9)
Protestant	7 (26.9)	25 (31.3)	17 (35.4)	15 (34.1)	64 (32.3)
None	8 (30.8)	12 (15.0)	3 (6.3)	10 (22.7)	33 (16.7)
Other	4 (15.4)	6 (7.5)	7 (14.6)	5 (11.4)	22 (11.1)
Education, *n* (%)					
School levels[b]	0 (0.0)	2 (2.5)	1 (2.1)	5 (11.1)	8 (4.0)
Post school education[c]	11 (42.3)	39 (48.1)	19 (39.6)	20 (44.4)	89 (44.5)
Tertiary level[d]	10 (38.5)	15 (30.9)	22 (45.8)	16 (35.6)	73 (36.5)
University degree	5 (19.2)	15 (18.5)	6 (12.5)	4 (8.9)	30 (15.0)
Employment status at death of the child					
Working	10 (38.5)	41 (50.6)	28 (58.3)	16 (35.6)	95 (47.5)
Off work[e]	16 (61.5)	40 (49.4)	20 (41.7)	29 (64.4)	105 (52.5)
Employment status at time of the survey					
Working	19 (73.1)	65 (80.2)	43 (89.6)	39 (86.7)	166 (83.0)
Off work[e]	7 (26.9)	16 (19.8)	5 (10.4)	6 (13.3)	34 (17.0)
Family income[f], *n* (%)	*n* = 19	*n* = 66	*n* = 44	*n* = 40	*N* = 169
≤ CHF 100,000.-	9 (47.4)	33 (50.0)	17 (38.6)	24 (60.0)	83 (49.1)
> CHF 101,000.-	10 (52.6)	33 (50.0)	27 (61.4)	16 (40.0)	86 (50.9)
Deceased child was the only child, *Yes* (%)	1 (3.8)	13 (16.0)	6 (12.5)	4 (8.9)	24 (12.0)
Previous loss of a child, *Yes* (%)	4 (15.4)	6 (7.4)	5 (10.4)	2 (4.4)	17 (8.5)
Place of death	***n* = 16**	***n* = 51**	***n* = 29**	***n* = 28**	***N* = 124**
Intensive care unit	10 (62.5)	47 (92.1)	13 (44.8)	5 (17.8)	75 (60.5)
Hospital	2 (12.5)	0 (0.0)	8 (27.6)	11 (39.3)	21 (16.9)
Home	3 (18.8)	3 (5.9)	6 (20.7)	11 (39.3)	23 (18.6)
Somewhere else	1 (6.2)	1 (2.0)	2 (6.9)	1 (3.6)	5 (4.0)
Deceased child's age					
in days, *Mdn* (*range*)		5 (1 – 26)			Na
in years, *Mdn* (*range*)	0.5 (0.1 – 9.1)		4.8 (0.1 – 17.2)	8.0 (1.7 – 17.4)	3.3 (0.1 – 17.4)

6.4.1. Parental experiences and perceived satisfaction with care

Overall parental experiences and their perceived satisfaction with care their child received will first be summarised, followed by more detailed reporting, focusing on differences between the diagnostic groups within the six quality domains. Parents rated experiences with their child's EOL care as generally positive (Figure 2). After accounting for the different scoring ranges among the six quality domain scales (7-point and 5-point), experience scores were highest for the domain *relief of pain and other symptoms* (*M* = 4.99, *SD* = 1.05) and lowest for the domain *continuity and coordination of care* (*M* = 4.29, *SD* = 1.37). Across all six domains, parents of children with cancer rated their experiences during their child's EOL care highest (*M* = 4.80, *SD* = 0.51), while parents of children with a neurological condition rated their overall experiences lowest (*M* = 4.51, *SD* = 0.44). The cardiology, neurology and oncology groups all showed the same pattern in experience scores across the six quality domains. Parents of neonates showed a different pattern, with a significantly lower score in the domain *shared decision making* (main effect, *P* = 0.001) and a high score in the domain *relief of pain and other symptoms* (*M* = 5.13, *SD* = 1.01).



Quality domain[a]	Parental experiences Overall *M* (*SD*)	Perceived satisfaction with care Overall *M* (*SD*) with an offset of -1
1. Support of the family unit	4.98 (1.01)	4.96 (1.48)
2. Communication	4.68 (1.06)	4.95 (1.42)
3. Shared decision making	4.82 (1.23)	5.07, (1.42)
4. Relief of pain and other symptoms	4.99 (1.05)	4.95 (1.34)
5. Continuity and coordination of care	4.29 (1.37)	4.69 (1.55)
6. Bereavement support	4.96 (1.18)	4.89 (1.40)

[b] Means for the continuity of care and bereavement support domain were scaled to a range of 0 to 6.

Fig 2. Parental experiences and satisfaction with care according to the six quality domains

Overall perceived satisfaction with the child's EOL care was also rated highly, with a mean of 5.92 (*SD* = 1.05) out of 7 across all quality domains and all diagnostic groups. However, the score patterns of the four diagnostic groups across the domains showed a different picture than for experiences. The domain *shared decision making* received the highest satisfaction rating (*M* = 6.07, *SD* = 1.42), and the domain *continuity and coordination of care* the lowest (*M* = 5.69, *SD* = 1.55). Consistently with parental experiences, parents from the oncology group rated their overall perceived satisfaction highest among all groups, while parents from the neurology group rated it lowest (Figure 2).

Support of the family unit

Parents or their dying child had access to a variety of support services. The most frequently reported were pastoral care (*n* = 108 Yes responses), followed by psychological care (*n* = 88) and physiotherapy (provided to the child, *n* = 71). Community, social and bereavement services were less common, but still offered to a quarter of the parents. Again, there were some differences between the diagnostic groups. Pastoral care and bereavement services were predominantly offered to parents of neonates (main effects, *P* = <0.008 and *P* = <0.007 respectively), and physiotherapy to children with a neurological condition (*P* = <0.001). Access to complementary medicine was mostly reported by parents with a child with cancer and almost never by parents of neonates. (*P* = <0.001). Thirty-five (18%) parents reported that they received specialised palliative care services. This was most often the case for parents of children with cancer (*n* = 7 Yes responses, 38%), and rarest for parents of neonates (6, 7%).

Communication in general and with physicians

Experiences with the clarity and honesty of the information physicians provided were analysed as single items and are summarised in Table 2. Parents from the oncology group consistently reported the most positive experiences (Table 2). All but seven parents (97%, *n* = 193) confirmed that they were informed that their child could die. However this occurred at differing time points, depending on the diagnostic group. In the cardiology and neonatology groups 42% (*n* = 11) and 36% (*n* = 28) of parents, respectively, reported being informed that their child would die prenatally (this response option was available for those two groups only). Information was provided within 24 hours or a few days prior to the child's death to an additional 59% (*n* = 46) of parents of neonates and 20% (*n* = 9) of parents in the neurology group. Most parents in the neurology and oncology group (76%, *n* = 35, and 89%, *n* = 40, respectively), and 42% (n = 11) of parents in the cardiology group were informed that their child was likely to die from a few months to more than six months before her/his death. Only the parents in the neurology and oncology groups were asked who had informed their child that she or he could die. In the neurology group most parents (91%, n = 42) reported that their child could not be informed because of the child's age or mental state. This was less frequently the case in the oncology group (32%, n = 14). Children with cancer were informed about the possibility of dying by their parents alone (27%, *n* = 12) or by their parents and a physician (25%, *n* = 11). A few parents from the oncology group reported that they did not want anyone to talk to their child about dying (7%, *n* = 3) or that their child did not want to talk about it (5%, *n* = 2).

Table 2 Communication domain:
Parental experiences related to clarity and honesty of information provided by physicians[a]

	Cardiology *M* (*SD*) *Mdn* (*range*)	**Neonatology** *M* (*SD*) *Mdn* (*range*)	**Neurology** *M* (*SD*) *Mdn* (*range*)	**Oncology** *M* (*SD*) *Mdn* (*range*)	**Total** *M* (*SD*) *Mdn* (*range*)
In general[b]					
Clarity,	5.88 (0.82) 6 (4 - 7)	6.00 (1.23) 6 (1 - 7)	5.83 (1.33) 6 (2 - 7)	6.23 (1.10) 6 (1 - 7)	5.99 (1.18) 6 (1 - 7)
Honesty	6.19 (1.10) 7 (4 – 7)	6.31 (1.20) 7 (1 – 7)	6.17 (1.26) 7 (2 – 7)	6.53 (0.84) 7 (3 – 7)	6.31 (1.13) 7 (1 - 7)
Treatment options to alleviate suffering					
Clarity[c]	6.08 (1.02) 6 (4 - 7)	5.96 (1.29) 6 (1 - 7)	5.79 (1.03) 6 (3 - 7)	6.31 (0.85) 7 (4 - 7)	6.02 (1.11) 6 (1 - 7)
Prospects of life-sustaining measures					
Clarity	6.14 (0.96) 6 (4 – 7)	6.11 (1.15) 6 (2 – 7)	6.37 (0.95) 7 (2 – 7)	6.53 (0.72) 7 (4 – 7)	6.27 (1.00) 7 (2 – 7)
Honesty	6.14 (1.35) 7 (2 – 7)	6.32 (1.09) 7 (2 – 7)	6.36 (1.23) 7 (1 – 7)	6.68 (0.62) 7 (5 – 7)	6.39 (1.09) 7 (1 – 7)

Note. No statistically significant differences between the diagnostic groups.
[a] Parents were asked to rate their experiences concerning communication with the attending physicians in terms of clarity and honesty. [b] All items were coded on a scale from 1 to 7. [c] Honesty response option not provided for this item.

Shared decision making

Overall, 60% of parents (*n* = 110) reported that a decision about resuscitating their child had been made. Those decisions were made significantly less often within the neonatology group (42%, *n* = 33), especially when compared with the neurology group (88%, *n* = 42; main effect, *P* = <0.001). Parents reported that resuscitation-related decisions were made by the family together with the health care team (HCT) in 39% of the cases (*n* = 45), by the family alone in 34% (*n* = 40), or by the HCT alone in 20% of the cases (*n* = 20). More parents in the neurology group reported that the decision was made by the family alone (52%, *n* = 22), whereas in the other groups it was commonly made by the family together with the HCT. Only parents of neonates were asked whether the cessation of life-sustaining interventions was discussed. Eighty-three percent (*n* = 63) confirmed that it was discussed and that the decision to end those measures was made by the family together with the HCT in 65% of cases (*n* = 46).

Relief of pain and other symptoms

Of all quality domains, experiences with pain management were rated most positively on a scale from 0 to 6 (*M* = 4.99; *SD* = 1.05), and highest of all by parents from the neonatology and oncology group (*M* = 5.13). Parents from the neurology group rated their experiences lowest (*M* = 4.63, *SD* = 1.17). Those parents were also less satisfied and reported the lowest value of all groups within this quality domain (Figure 2).

Parents were asked to rank the three symptoms from which their child suffered and that were most stressful to them from a list of more than 10 different symptoms. Problems with breathing was ranked most frequently by parents from the cardiology, neonatology and neurology groups, followed by pain. Parents in the oncology group ranked pain first. Breathing problems and pain were overall the symptoms most frequently ranked among the top three as being most stressful for parents. Other stressful symptoms were different according the diagnostic group. For the cardiology group, agitation and anxiety were frequently ranked in first or second place. Parents of neonates frequently ranked circulatory problems in first and third place among the top three. Parents from the neurology group frequently ranked mucus in the airways and seizures among the top three, and for parents from the oncology group, their child's fatigue and impaired verbal or nonverbal communication frequently appeared among the top three.

Continuity and coordination of care

Experiences related to continuity and coordination of care were rated least positively of all six quality domains by all parents. Between the diagnostic groups, parents of children with cancer rated their experiences highest, with a mean of 4.10 (*SD* = 0.81) on a scale from 1 to 5. This was also reflected in the overall satisfaction rating of that domain (on a scale from 1 to 7), where parents from the oncology group were more satisfied (*M* = 6.09, *SD* = 1.20) than parents from the other groups (*M* = 5.53, *SD* = 1.64) (Figure 2).

Parents from the cardiology, neurology and oncology group were asked who mainly supported them professionally in the organisation of their child's care. The most frequent answer for all groups was: a hospital-based physician (36%, *n* = 37). For the oncology group this was followed by a hospital-based nurse (26%, *n* = 11), which was less often the case for the neurology group (8%, *n* = 3). Evenly spread across all diagnostic groups, parents also reported that a community-based nurse supported them in organising care (18%, *n* = 19) but 19% (n = 20) answered that no one supported them. Main support was provided by a primary care paediatrician (PCP) in 8% of cases (*n* = 8).

Bereavement support

Sixty-nine percent (*n* = 137) of the parents stayed in contact with someone from the HCT shortly after their child's death. This applied most often to parents from the oncology group, with only 16% (*n* = 7) of parents having no further contact. Overall 65% (*n* = 130) of the parents reported having a follow-up talk with someone from the HCT. The lowest rate was reported by parents from the neurology group, where it was the case in 50% (*n* = 23) of respondents. Parents were asked to write down what kind of support services they used or still use during their bereavement. We received written information from 140 parents. Of those, 59 (42%) parents reported making use of psychological support services, followed by support groups with other bereaved parents (32%, *n* = 45). Other common answers were related to alternative support services (26%, *n* = 36) such as kinesiology, art therapy, dream therapy, and spiritual services (24%, *n* = 34) with a pastor or in a religious community.

6.4.2. Positive and negative experiences and quality of life

Parents were asked to describe three positive and three negative experiences related to their child's EOL care. Responses were classified according to the questionnaire's six quality domains and are summarized with frequencies and a sample quote in Table 3. Both positive and negative experience descriptions were most frequently about the support the family received.

Table 3 Positive and negative experiences

	Number of quotes[a]	Sample quote
Positive experiences	**180**	
Support of the family unit	174	*Our individual needs were always supported. (18:6)*[b]
Communication	54	*Honesty when informing about our child's situation. (22:2)*
Shared decision making	8	*Ethics council helped to take the right decision. (35:4)*
Relief of pain and other symptoms	8	*Oxygen support at home, mail order of medication. (101:2)*
Continuity and coordination of care	46	*Reachability day and night (hospital and community care). (17:1)*
Bereavement support	39	*That a lot of time was provided* (by the hospital) *to be with my son after his death. (3:1)*
Negative experiences	**165**	
Support of the family unit	110	*I felt left alone (75:3)*
Communication	73	*Not having enough information about my child's situation (61:3)*
Shared decision making	14	*The night doctors did not support and follow our decision to end treatments. (5:7)*
Relief of pain and other symptoms	27	*Pain and shortness of breath. There was a phase when effective medication lagged behind the symptoms. (33:10)*
Continuity and coordination of care	53	*When the physicians and nurses always change. (116:2)*
Bereavement support	51	*No follow up care for us after her death. I needed to find my own psychologist/support group. (97:7)*

[a] Each listing of a positive or negative experience (quote) was potentially coded to more than one domain. [b] Numbers in parenthesis represent the quote's identification number.

Current quality of life (QoL) was rated high overall, with a mean of 7.19 (*SD* = 2.09) on a scale from 0 to 10. However six parents reported low QoL below the 5th percentile with values between 2.30 and 0. Parents who had lost a child due to cancer rated their QoL lower than parents from the other groups with a mean of 6.55 (*SD* = 2.17) vs. cardiology (*M* = 8.09; *SD* = 1.18), neonatology (*M* = 7.46; *SD* = 1.88), and neurology (*M* = 6.84; *SD* = 2.49). To further investigate this difference and based on the sample's characteristics, the parents' age and income was added to the GEE model. In this extended model, income was the most influential predictor of QoL ($P = 0.002$) with higher income predicting higher QoL.

Finances were also one of seven response options for the question about areas of the parents' personal life that were negatively influenced by the illness and death of their child (Figure 3). Overall, 37 (18.5%) parents responded that finances were an area that was negatively influenced. Significantly fewer parents in the neonatal group reported that their finances were negatively affected compared to the other groups (main effect, $P = <0.001$) whereas in parents from the neurology group the proportion of "Yes" responses was highest (35%). The areas most commonly affected across all diagnostic groups were their own health ($n = 80$, 40%) followed by the family ($n = 70$, 35%) and the partnership ($n = 64$, 32%) (Figure 3).

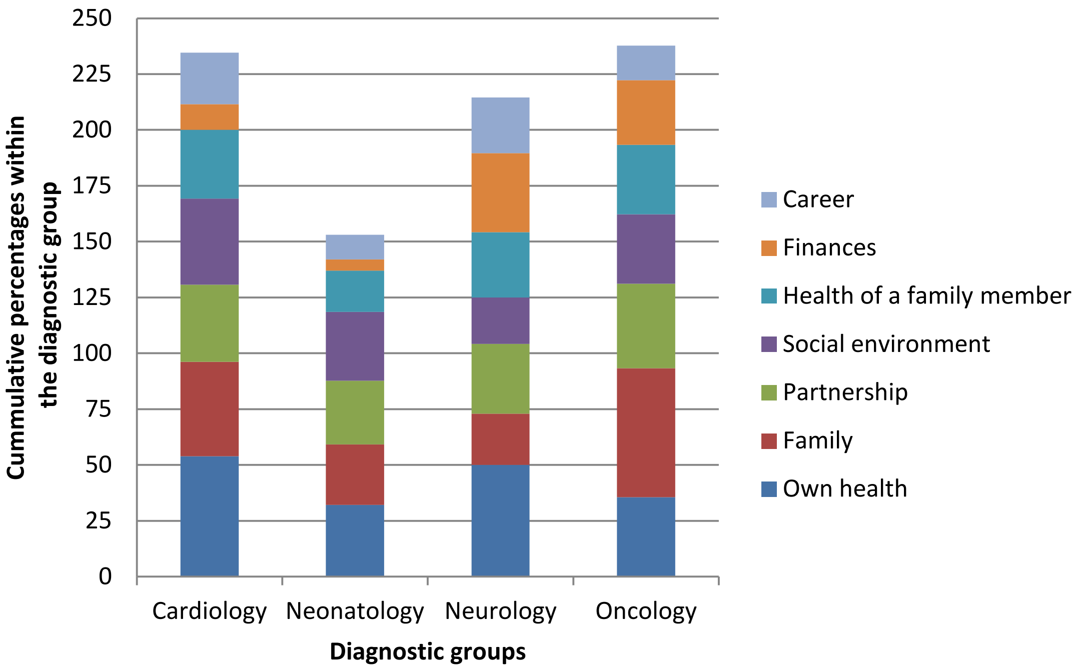


Fig. 3 What areas of your personal life were negatively influenced by the illness and death of your child?
Parents could choose from a list of seven potential areas and check all that applied. The areas are represented by the different colours. The number of checks per participant was for cardiology 2.3, neonatology 1.5, neurology 2.3, and oncology 2.2.

6.5. Discussion

This is one of the few studies that quantitatively described and explored parental experiences related to their child's EOL care in a population-based sample of bereaved mothers and fathers of children from the major diagnostic groups in which childhood deaths occur. This allowed us to compare findings in four distinct diagnostic groups, which adds to existing knowledge about parental perspectives. Overall, parental scores on their experiences and perceived satisfaction with their child's end of life care were high across all six quality domains. Parents of a child with cancer rated their experiences highest in most of the six quality domains and reported the highest satisfaction with care. The lowest scores were mainly reported by parents from the neurology group, with the exception of the domain *shared decision making*, where parents of neonates reported significantly less positive experiences.

6.5.1. Satisfaction with care

Evaluation of health care is considered the most important purpose of measuring patient/parental satisfaction [27]. However, ceiling effects are a methodological concern when measuring the construct of satisfaction, i.e. high levels of satisfaction are consistently reported, which reduces the ability to discern differences. Such high levels might be due to inherently low expectations and should thus not be automatically interpreted to mean that care was good but simply that nothing bad happened [27]. This was, for example, the case in Wolfe et al.'s study [28] which showed substantial parent-reported EOL suffering in children with cancer as well as, simultaneously, high levels of satisfaction with care. A similar mechanism might contribute to this study's high perceived satisfaction levels, e.g. results for the domain *shared-decision making* of the neonatology group. Assessing specific experiences with aspects of care shows promise as a means of overcoming limitations in general satisfaction measures. This is supported in this study where there were more variable results between the six quality domains and four diagnostic groups, and substantial differences were present in regard to some domains. The role of domains is important as they present a structural framework for good quality care and evaluating experiences offers insight into processes of care that is less biased by expectations than measuring satisfaction of care. The domains chosen for the PaPEQU were established by experts in the field and mostly in accordance with domains established through exploratory factor analysis by Widger et al. [29]. Future research however, should further focus on empirical model development and testing to measure good quality paediatric EOL care [19].

6.5.2. Communication

Parents consider genuine communication with sincere and honest provision of information to be most central [3], and lacking or poor communication were recurrent themes in the Aschenbrenner et al. [2] review and a recent meta-synthesis by Xafis et al. [30]. Experiences and satisfaction scores related to communication in this study were high. However, extreme negative outliers were present and reflected in the parents' written comments about their negative experiences, where a lack of or inconsistent provision of information and also insensitive communication in general were predominantly described. Similar complaints were also described in a meta-synthesis on the information needs of parents facing an EOL decision for their child [30]. Even though it might affect only a few parents, these experiences should not be taken lightly as their negative impact can last for years after the traumatic event.

6.5.3. Shared decision making

Consistent with our findings, a majority of paediatric deaths occur in the ICU [31], and, especially for neonates, these are preceded by a decision to limit or withdraw life-sustaining measures [32]. More than 80% of parents from the neonatal group in this study reported that the withdrawal of life-sustaining treatment had been discussed; however only 65% indicated that they made those decisions in consultation with the HCT. Some parents felt that the decision was made by them alone or by the HCT alone. This finding contradicts what was found in a another Swiss study evaluating how EOL decisions were made in neonatology and how consistently a framework for structured ethical decision making was applied [33]. They concluded that 92% of parents were actively involved in the decision by having received full information about the baby's condition, prognosis, therapeutic possibilities and the approach the treating team would take [33]. A Canadian study exploring processes of death in neonates reported that there was agreement between physicians and parents in 84% of cases where a decision to withdraw life-sustaining measures was made [32]. All parents in our study were also asked if there had been a decision concerning the potential need to resuscitate their child; 60% said there had been. However, one third of those parents, predominantly parents from the neurology group, indicated that they perceived themselves making this decision without input from the HCT. Such results are unexpected and call into question our perceived reality of family-centred care. The child's best interest is always central in making these decisions. However, professional caregivers and parents have their own personal perceptions, values and interpretations of what is best for the child and the balance of power is not equal in this context. Following the traditional principles of bioethics may not ensure that parents have the opportunity to participate to their satisfaction in those important decisions. It might be that other communication models ensuring shared decision making should be considered and introduced. One such model, communicative ethics, is explained and discussed in the context of neonatal intensive care by Daboval and Shidler et al. [34]. It builds on the shared-decision model which is considered as gold standard [35], acknowledging that the decision made cannot be separated by the communicational process used to reach it [34].

6.5.4. Relief of pain and other symptoms

Although experiences with alleviation of suffering were rated highest among the six quality domains and perceived satisfaction levels second highest, parents still reported that their child experienced a wide variety of stressful symptoms, indicating that significant symptom burden is present at EOL [36]. Breathing changes are part of the dying process and were probably witnessed by most parents. Even though it is recommended that parents be informed about what physical changes to expect when their child is dying, witnessing this process remains very stressful. In a US study with 50 bereaved parents of children with cardiac diagnoses, breathing difficulties were associated with considerable suffering in 77% of the 30 parents who reported that symptom [15]. Pain was another frequently reported symptom in our study and experiences related to pain management were rated lowest by parents from the neurology group. This reflects the tremendous challenges we face when caring for nonverbal children with a variety of neurological impairments [37].

6.5.5. Continuity and coordination of care

Continuity and coordination of care is recognized as an important factor in promoting caring, reducing parental frustration, and enhancing parents' confidence in the quality of their children's care [38]. Experiences as well as perceived satisfaction with this aspect of care were rated lowest of all domains by parents in this study and point to an area with a need for substantial improvement. Supported transition between inpatient, outpatient and home care is essential to high quality EOL care [4]. This however, requires appropriate structures concerning health care services and professional palliative care support. While parents in our study felt most supported by a hospital-based physician or nurse, many felt left alone. Community-based nurses and PCPs only played a minor role. The latter was also described in a study that explored the involvement of PCPs when their patients faced the EOL [39]. The fact that multidisciplinary PPC teams only exist in three Swiss paediatric hospitals and only 18% percent of all parents reported having received specialised PPC services might contribute to our study's findings.

6.5.6. Bereavement support

Continuity and attention remain important after the death of the child. Parents described great appreciation for staff sending cards or attending the child's funeral and said that the loss of this connection added to their bereavement [38]. Interpreting our study's findings, this might be especially true for parents of a child with cancer who, although the palliative care period tends to be rather short, experience a long illness/treatment phase with many hospital stays in a dedicated paediatric oncology unit, creating a special bond between the family and the HCT [40]. Only half of our parents from the neurology group reported having had a follow-up consultation with the former treating team/physician. We cannot conclude whether this reflects those parents' wishes or rather highlights an area for improvement. Several other studies, however, indicate that parents value the HCT following up with them, that they need help in preparing for what to expect at the time of death, including funeral arrangements, and that they want bereavement services to be available immediately after their child's death [4]. As also reported by the parents in our study, many desire contact and peer support from other families who have lost a child [40].

6.5.7. Positive and negative experiences and quality of life

The death of a child disrupts the parents' well-being and can influence various areas of their personal lives and their quality of life (QoL) [41]. The perception of the quality of medical care has been described as a factor associated with psychosocial morbidity in parents who have lost a child due to cancer [42]. The parents from the oncology group in this study reported the most positive experiences and the highest perceived satisfaction with care. The finding that they reported the lowest current QoL of diagnostic groups was therefore surprising. A similar result was found by Bergstraesser et al. [43] in their study of dyadic coping of parents, where mothers from the oncology group had poorer psychosocial health than fathers and parents from other diagnostic groups. Further exploration of our results revealed that income appeared to be the most influential predictor of QoL. This could in part explain the aforementioned finding, since parents from the oncology group were more frequently in the lower income category. Interestingly, financial strain was described as a major burden for families of children with life-threatening illnesses in Western Australia [14]. The study reported that families in the non-cancer group experienced a high degree of financial strain, which is congruent with our findings that the neurology group reported finances as an area negatively influenced more often than did parents from the other diagnostic groups. Although expenses in this patient group are mostly covered by the Swiss Federal Invalidity Insurance, refund processes seem to work very slowly and putting parents in an economical burdening situation. In addition, financials strains in these families are also due to the duration of care and the fact that one parent will be fully absorbed by the task of care, leading to loss of earning.

6.5.8. Limitations

Although a high percentage of parents completed the questionnaire, generalisability might still be limited as only parents who had previously provided informed consent received the questionnaire. Our participation rate of 51% lies within a wide participation range from below 20% to up to 80% reported in other studies with bereaved parents [1, 29, 44]. Our findings might be biased in that only parents with favourable experiences may have been motivated to participate. Also, the requirement of being proficient in German, French or Italian excluded some migrant residents representing cultural minorities. The retrospective nature of this study could have introduced a recall bias and parental perceptions of care could have changed over time. However, during the cognitive testing phase of questionnaire development, remembering details of the devastating experience of losing a child was not a problem for participants [19].

6.6. Conclusions

Our findings show positive parental experiences of their child's EOL care and high perceived satisfaction with the care their child received. In the context of this national study with heterogeneous inpatient and community care settings the differences between the four diagnostic groups were small and within one scoring category. Nevertheless there are some areas worthy of our attention. Parents of neonates reported significantly lower experience ratings related to shared decision-making. As these parents mostly face a decision to withdraw life-sustaining measures, particular attention should be paid to shared decision-making processes. Apparent consensus between the parents and the HCT does not imply that the process was well perceived by the parents. Differences as to how discussions went and on the opportunities or time available might be present.

Parents of children with neurological impairments face many challenges. Symptom management can be a source of distress for parents, as the children are mostly nonverbal and the potential for suffering is high due to a variety of impairments. This makes them dependent on a variety of different care services, which creates a highly complex care environment with a great need for continuity and coordination. Experiences with continuity and coordination of care were rated lowest of all quality domains by parents from all diagnostic groups, and perceived satisfaction with care within this domain was lowest as well. This might be the direct result of lacking specialised PPC services. We have to recognize that the integration of specialised PPC has the potential to minimise lost opportunities for supporting and assisting parents. This has been acknowledged by policy in many countries and there is a growing availability of specialised PPC programs worldwide; unfortunately Switzerland lags behind in this area.

However, the development and implementation of needs-driven and specialised services will fall behind if the benefits of these services are not evidenced in the near future. Structural evaluation and performance data provide one part of the evaluation. However intervention research is needed evaluating processes and outcomes that are meaningful to patients and their families, siblings included. Promoting the best possible outcomes after such a devastating experience has implications for the whole family, the healthcare system and society.

6.7. Competing interests

The authors declare that they have no competing interests.

6.8. Author's contributions

EC and EB designed the study. KZ elaborated the methodology, developed the questionnaire and conducted the study under the guidance of EC and EB, had full access to the data and performed the statistical analysis. AS, PF and CG were involved in recruitment processes. KM contributed to the qualitative data analysis. All authors contributed to the manuscript and approved the final version.

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Chapter 7
Synthesis and Discussion

How to best care for children at their EOL and provide support for their families, which have to endure a most painful event that is considered to create a ripple effect of grief for the rest of their lives [1]? There is no easy and simple answer to this question and with answering one, more questions arise. This dissertation project, as part of the nation-wide PELICAN study, provides comprehensive and direly needed information about the characteristics and circumstances of childhood deaths due to a life-limiting CCC or prematurity in Switzerland. It allows invaluable insight into the experiences of parents and produced a promising instrument for future use in initiatives to improve paediatric EOL care in heterogeneous healthcare settings internationally.

7.1. Key findings

In a population-based sample of 149 children including prematurely born babies, 62% died in an ICU, 84% of them following the withdrawal of life-sustaining treatment. Only 17% of the children died at home and 48% (n = 72) spent at least one day at home during the last four weeks of their life. Half of those 72 children received professional community care services. EOL care was characterized by a variety of prevalent symptoms, on average 6.42 symptoms per patient, and intensive treatments with high medication counts. Parents rated their experiences with their child's EOL care mostly positive, with high satisfaction with care. Among the six quality domains that structured the questionnaire, continuity and coordination of care was rated lowest in regard of experiences and satisfaction with care as well. Relevant differences between the diagnostic groups could be demonstrated and were most accentuated between the neonatology group and the other groups.

7.2. Parental experiences, needs and satisfaction with care as measures of quality of care

Quality of care is a complex construct and can be assessed on different levels within the healthcare system and from different perspectives. Information from which inferences about care quality can be drawn were most famously conceptualised into the tree categories of structure, process and outcome by Donabedian et al. [2]. The expression of satisfaction or dissatisfaction represents the patients' judgement on care and is considered by some as an outcome measure of the care received [3, 4]. Although, Donabedian et al. [2] stated that satisfaction with care concerned mostly interpersonal processes as part of practitioner's performance in delivering health care and is therefore a measure of the process of care. Satisfaction with care, including the perspective of relatives, might be even more important in the field of palliative and EOL care, where outcomes such as mortality, recovery or readmission rates are inappropriate [5]. However, confusion between patients' satisfaction and the actual care experiences exists and these concepts are often used interchangeably [6]. Additionally, ceiling effects, i.e. high satisfaction with care ratings that reduce the ability to discern differences, are an inherent problem when measuring satisfaction with care [5].

7.2.1. Parental experiences and satisfaction with care

As discussed in chapter 6, parental experiences as measured by asking specific questions organised in scales with an adjectival response option showed more variable results within and between the six quality domains as the corresponding single satisfaction question for each domain. Also, the pattern of responses was not consistent between experience scores and satisfaction scores. Therefore it seems that parental experiences and satisfaction with care might be two distinct constructs. This assumption was recently supported by a series of studies as part of a doctoral thesis covering patients' perceptions and satisfaction of care in hospitals [6]. A lack of theoretical underpinning in the measurement of satisfaction in healthcare was recognised. Nevertheless, a relationship between satisfaction and the fulfilment of expectations has often been hypothesised, meaning that one is satisfied when he gets what he expects [5, 7]. If this assumption held true, high satisfaction levels with little variance should be expected when evaluating paediatric EOL care as families' expectations during this most stressful phase might be inherently low. Additionally it could then also be questioned whether satisfaction with care really measures quality of care or whether it is not rather just an indication of better experiences than expected [5]. The question then arises of what influences patients' satisfaction with care? When used as a construct to assess quality of care, it is important that the services received rather than external factors, i.e. socio-demographics, determine levels of satisfaction. In our study we did not explore the relationship between socio-demographic factors such as age, gender, educational level of participants, or family income. These factors however, have been studied in the past with inconclusive results [5, 8] and it remains uncertain what factors mainly determine satisfaction with care. In that sense, assessing parental experiences in the context of paediatric palliative and EOL care, and possibly even in general, seems to be a valid and appropriate approach to measure the quality of care processes.

7.2.2. Parental needs

In addition to the quantitative assessment of parental experiences we strived to integrate specific closed questions concerning parental needs. In our conceptualisation of the PaPEQu we theorized that specific parental needs related to their child's EOL care would emerge based on how they experienced the professional care they received. This is an opposite view on needs than for example described in Maslow's hierarchy of human needs [9], where needs are inherently present. Following our mental framework, we wanted to assess the importance of needs as a result of experiences made. This led to the formulation of items asking about the importance of specific needs in the conditional mood. Importance ratings assessing parental needs have been used in paediatric research before. The "needs of parents of hospitalised children" (NPQ) is an instrument developed by Kristjánsdóttir [10] that has been used in different countries and languages [11]. The comprised needs statements used a two-part item response format rating the importance and fulfilment of needs [10]. The same response format was also taken up by Meert et al. [12] for their BPNA questionnaire that assesses parental needs and fulfilment around the time of their child's death in the PICU.

Our results of the needs-related items in the PaPEQu, assessing importance of needs parents would have had based on their experiences made during and after the EOL of their child, showed high ceiling effects and little variation across the four diagnostic groups. Similar effects were found when applying the NPQ questionnaire as none of the needs statements were rated as unimportant [10]. The high importance ratings and the fact that we did not assess to what extent the needs were met limited the interpretation of the results of this part of the PaPEQu. It became apparent that healthcare provided not only determines what needs are expressed but also to what extent those needs are met [13].

7.2.3. Reflections on the Parental PELICAN Questionnaire (PaPEQu)

Initial validity of the six evidence-based quality domains that structured the PaPEQu was demonstrated in our instrument deveopment study. This study's results were partly confirmed by the testing results of another recently developed instrument to measure the quality of children's EOL care [14]. The six subscales that initially structured that other, similar instrument overlapped with five of the six PaPEQu domains: *support of the family unit, communication, shared decision making, relief of pain and other symptoms, and bereavement support (continuity and coordination of care* missing*)*. Exploratory factor analysis during their phase two of instrument development revealed four additional domains related to individual support to the child and siblings, and to information sharing among health professionals and structures of care [14]. One important difference to the PaPEQu that stands out is the integration of the siblings' perspective, even though not self-reported but as reported by mothers. There is no question that special attention is required to meet the needs of siblings of a dying child and a few issues related to that will be covered in paragraph 7.3.

A major weakness of the PaPEQu lies in our operationalisation of parental needs. Assessing only the importance of needs derived from experiences does not capture the needs construct in its entirety and in the context of healthcare evaluation, assessing the level of need fulfilment may represent the quality of services provided.

Although the PaPEQu will need some revisions for future use, the instrument shows promise in moving forward towards a valid measure of processes of paediatric EOL care. The burden of participating in a questionnaire survey should not be underestimated for bereaved parents and professional support should be offered for anyone in need. The questionnaire response rate of 89% of parents that consented to participate beforehand however, is an indication that parents are motivated to contribute.

7.3. Siblings

Siblings are often referred to as “shadow children” as they tend to become invisible as a consequence of their parents’ enormous investment in caring for the ill child. The effects on psychological well-being and functioning in siblings of children with CCC have been summarised regularly in meta-analyses and integrative reviews and the conclusion is that those siblings were more at risk for psychological problems [15-17]. Obviously this evidence is based in the field of psychology; a circumstance that holds true for most existing literature around (bereaved) siblings. Clinical (nursing) research on bereaved siblings is extremely scarce. However, certain issues related to siblings of a dying child have been described in various books [18-20]. How children perceive and understand the illness trajectory and -experience of their dying sibling and the parents’ reactions, hugely depends on their own age, i.e. developmental stage [21]. Helping small children “understand” is challenging and often perceived as futile. However, creating confusion and actions of ignorance add to the feeling of “hurting” in the sibling. Parents and healthcare staff also are often reluctant to involve the sibling(s) into the dying child’s care, trying to protect them from trauma. Unfortunately this reinforces the siblings’ feelings of “not belonging” and “not being enough” [20]. Siblings who lost a sister or brother due to cancer - as participants in a qualitative study - expressed their dissatisfaction concerning information and support they received and their involvement in the dying process [22]. Those feelings of abandonment can persist long into bereavement and it is important that the needs of siblings are recognized and support is offered [18].

In striving to provide holistic and good quality family-centred paediatric EOL care it seems apparent that the siblings’ perspective should be part of care evaluation. If not, we continue denying them the opportunity to voice their experiences and a piece is missing towards a family systems perspective. However, including children in research opens a whole new dimension of ethical considerations which are challenging to address. It also remains unclear whether parents would be willing to let their healthy children participate in research projects. A recent UK study to explore parents’ and young people’s perspectives of hospice support, including siblings aged eight years and above, achieved approval from the ethics committee. In the end however, the researchers were unable to recruit any siblings to take part in the study. Unfortunately, no explanation was provided [23].

7.4. Paediatric EOL care in distinct diagnostic groups

Based on diagnoses mainly responsible for childhood death due to a life-limiting CCC or prematurity we classified the PELICAN study patients into four distinct diagnostic groups: cardiology, neonatology, neurology, and oncology. The neonatology group comprised all newborns between 1 and 28 days of age, independent of their medical diagnoses. The neurology group was the most heterogeneous, including patients with endocrine, nutritional, metabolic diseases and diseases of the nervous system, and congenital, chromosomal conditions. Comparisons between these four diagnostic groups in regard to parental experiences and care practices highlighted relevant differences adding to the body of scientific knowledge.

7.4.1. Cardiology

Children with congenital heart conditions and diseases of the circulatory system represented with 19 patients (13%) the smallest of the four diagnostic groups in the PELICAN study. This small number however is biased by the fact that all neonates that died within their first four first weeks of life, were assigned to the neonatology group per study definition. Five patients in the neonatology group suffered from a congenital heart disease.

EOL care in the cardiology group was characterised by a high dependency on intensive care; more than half of the patients received medical treatment requiring anaesthesia, mostly for surgery, 37% needed cardiopulmonary resuscitation at some point and 21% were dependent on extracorporeal membrane oxygenation (ECMO). Medication count in the last week of life was highest compared to all other diagnostic groups. This can be explained by the higher rate of cardiopulmonary resuscitation in this group compared to the other groups, which is associated with a long list of as needed drug orders. Consequently, 67% of the patients died in a PICU. Comparable evidence about practices at EOL for children with cardiac disease is rare. Only one study was found that looked at patterns of care at end of life in children with advanced heart disease [24]. Accounting for the different inclusion criteria in that study where only children who died in the hospital were included, that study's results were very consistent with ours [24].

The illness trajectory and progression of paediatric advanced heart disease is highly variable and often unpredictable. With advancements in surgical techniques and the broadening of treatment options the impression might form that "broken" hearts can always be fixed. Timely initiation of PPC is challenging in this context. Rather than finding the "right moment" during the hectic phase of a declining health status, PPC should be an integral component from the moment on a child is diagnosed with a complex congenital heart disease. The PELICAN I study showed that especially for children in the cardiology group, this is often the case already before birth. Through a recent review about palliative care models in the perinatal setting it has been recognised that the antenatal period should also be considered [25]. The authors found evidence that antenatal palliative care have been explicitly organised in PPC programmes in some cases and that the concept was mentioned in 27 of the 101 articles included in their review. They concluded that antenatal palliative care is the latest advancement in the conceptual evolution of perinatal palliative care that started with pain relief and evolved from family-centred to integrative care during the last decades [25].

Synthesising the scarce evidence about the characteristics and circumstances of death in children with severe cardiac problems it seems obvious that considerable suffering is involved. The association between high intensity care and suffering has been suggested in children with cancer at their EOL [26]. It would be important then that PPC were an integral part of the treatment concept in children with congenital heart disease, starting early on and also in cases where the possibility of a "curative" surgery exists.

7.4.2. Neonates

Neonates constituted the largest group in our study which is in accordance with what is already known about conditions and diseases responsible for childhood death [27]. This patient population was different from the other diagnostic groups on several levels; time between diagnosis and death was with several days the shortest by far, almost all neonates died in an ICU, were mechanically ventilated and most relevantly, death was preceded by a decision to withdraw life-sustaining interventions in 91% of the cases. Making those decisions is challenging for the HCT involved and certainly most difficult for parents who might feel that they have to decide over life and death of their child during a phase of time that is characterised by immense vulnerability and uncertainty. Parents in the PELICAN II study rated their experiences with shared-decision making significantly lower than the parents from the other diagnostic groups. However their satisfaction with care rating for that quality domain was not below the whole group's average. A circumstance that might best be explained with the presence of low expectations as discussed in chapter 6.

Possible factors that affect parental decision-making for medically complex infants have been synthesised in a recent integrated literature review including 31 articles from the years 2000 to 2013. One major theme of factors influencing the parents' ability to make decisions was "communication" including information needs and the delivery of needed information [28]. The need for compassionate communication, building on trust between the family and the healthcare team is well known and is a main component in the framework of good quality paediatric EOL care that structured the PaPEQu. The concept of shared-decision making incorporates communication processes between the HCT and the family, and builds on a trustworthy relationship. Contrary to other models of decision-making, information sharing between the healthcare professional and the family is a continuous mutual process. Both the professionals and the family bring in their own expertise, experiences and attitudes and decisions are made in partnership [29]. Whether the shared-decision model could be implemented in neonatology has been reflected upon in the past, with the conclusion that the attempt to achieve a joint decision should be made [30]. Several studies reported on high percentages of active parent involvement in decision-making in the NICU setting [31-33]. Although it has been described that ethical guidelines regarding the withdrawal of intensive care are being followed [32], this does not automatically imply that shared-decision making as a process of care was applied. It therefore remains unclear to what extent the concept of shared-decision making is integrated in neonatal palliative care in Switzerland and how this might be associated with the experiences of parents from neonates in the PELICAN II study.

Further exploration of possible factors influencing parental decision-making is indicated to explain the significantly lower experience ratings found in parents of neonates compared to the other parents participating in the PELICAN II study. Other factors than the ones related to "communication" that were described in the abovementioned review were: factors related to the child's condition and the availability of treatment options, the child's best interest, religiosity and spirituality, and parental factors such as sociodemographics [28]. As some of this information is available from the PELICAN datasets, future secondary data analysis could potentially add further insight on this issue.

7.4.3. Neurology

The primary diagnoses of the patients in the neurology group were more diverse than in the other diagnostic groups. Most of the represented diagnoses affect the central nervous system leading to physically and cognitively severely impaired children with many distressing symptoms [34]. For most of those conditions cure is not possible, disease progression is variable and the decline in health can by slow. Life-threatening health exacerbations are part of the illness trajectory and it is often not possible to know when the next event might end in the death of the child [34]. Apart from the "continuity and coordination with care" quality domain, parents of children in the neurology group rated their EOL care experiences consistently lower compared to the other parents and their satisfaction with care was generally lower as well. They also reported more often than all other parents, that their family's financial situation was negatively influence by the child's illness and death. The long illness duration seen in children with neurological conditions usually leading to the full absorption of one parent and together with rigid and slow insurance refund processes make those families especially susceptible for financial strains, constituting a problem that has been described outside of Switzerland as well [35].

Based on these patient's population illness trajectories with high care dependency over longer periods of time, families of a child with a life-limiting neurologic condition may benefit tremendously from PPC services. Benefits of PPC specific to this group of patients were synthesized recently and included fewer invasive interventions at the EOL, decreased hospital length of stay, and improved symptom management [34]. Symptom control to establish comfort in these patients is challenging as they suffer commonly from neurologic symptoms such as spasticity and dystonia [36]. These type of symptoms lead to significant physical discomfort, i.e. pain, which is probably under-recognized and – treated as these children are commonly not able to express their discomforts verbally [37]. Spasticity/dystonia was more frequently documented in children from the neurology group compared to the other diagnostic groups in the PELICAN I study. However, the prevalence of 28% was not high and an under-documentation of these symptoms should be assumed. Interdisciplinary teamwork including the expertise of the child's parents is highly recommended in managing the complex symptomatology of children with severe neurologic impairment [36].

Many of the other discussed benefits and also barriers from the Hauer & Wolfe article [34] are transferable to our entire study population and the relevant issues will be discussed under paragraph 7.5. about the provision of paediatric EOL care in Switzerland. To conclude the specificities of the neurologic group, it can be said that the variety of primary diagnoses leads to the involvement of different specialists and healthcare providers. When these providers work independently, care can be fragmented, adding to the family's burden. Additionally because the care is complex and very challenging, healthcare professionals may reach the limit of their expertise which might negatively influence the validity of and certainty with end-of-life decisions.

7.4.4. Oncology

Experiences and satisfaction with care were rated consistently highest by parents of a child that died due to cancer compared to parents from the other diagnostic groups. One explanation might be that the care experiences from parents in the oncology group were positively influenced by components of standard paediatric oncology care that are also typical in the PPC model. In Switzerland, paediatric oncology care is provided in dedicated units situated in tertiary care centres and specialised children's hospitals. Just as in PPC, care is provided by a specialised multiprofessional team including social workers and psychologists. The provision of in-patient care by those dedicated teams creates an environment of second home for families and the perceived level of support is high. Outpatient clinic services are sometimes directly linked to the inpatient unit and even when separated, the teams collaborate. Through this mode of care delivery, fragmentation of care is much less an issue than, e.g, for families of a child with a neurologic condition.

What has just been discussed stands in contrast to the lower QOL found in parents from the oncology group compared to the other parents. There is no reported evidence about comparisons between parents of children with cancer and other bereaved parents. Factors associated with psychosocial morbidity among bereaved parents of children with cancer however, have been summarized in a systematic review including 13 studies presenting results mainly from a Swedish registry sample (5 studies) and an Australian database sample (2 studies) [38]. Those factors included economic hardship, duration and intensity of the child's cancer-therapy, location of death and the perception of medical care [38]. Economic hardship, i.e. lower income, was the most influential predictor for lower QOL in our exploratory model, but our insight into determinants of QOL in bereaved parents with cancer is limited as it was not one of the PELICAN study's research question.

QOL certainly is an important outcome in bereaved parents in general and further conceptual work exploring possible influencing factors including characteristics of EOL care, the parents' perspectives and socio-demographic factors is indicated.

7.5. The provision of paediatric EOL care in Switzerland

The overall positive experience ratings and high satisfaction with care reported by parents in the PELICAN study seem to indicate that all is well. However, on the single item level, extremely negative responses were present including low satisfaction and in six cases QOL ratings below the 5th percentile. Examples from the listings of negative parental experiences showed that deficiencies in all of the six quality domains existed and we were concerned by the implication of some of the statements. Switzerland lags behind in the movement of PPC programme development seen internationally over the last decade. In the year 2015, the "Schweizerisches Institut für ärztliche Weiter- und Fortbildung, FMH"acknowledged and promoted palliative care as a new sub-specialty within the field of medicine and professorships exist at two Swiss universities. So far, only two of the five existing Swiss university children's hospitals provide PPC as part of a programme with a specialised multidisciplinary team. It should be expected that university hospitals offer the whole range of subspecialty healthcare services, so hopefully more PPC programmes will be implemented in the future. This current lack of specialised PPC is reflected in some of the PELICAN I and II results which will be discussed hereinafter.

7.5.1. Place of death

Sixty-two percent of the PELICAN study patients died in an ICU. A similar high rate of 65% was found in a sample of 220 children that died between 2008 and 2010 and were recruited by a Canadian university-affiliated hospital [39]. On the one hand, this high rate can be explained by the high proportion of neonates and NICU deaths in our and the Canadian study's sample. Neonates, especially when born preterm, are highly dependent on intensive treatment, e.g. mechanical ventilation. When life-sustaining treatment is withdrawn, time until death occurs is mostly short with minutes to a few hours (unpublished data from the PELICAN ICU sub-study). Under those circumstances, there is no other realistic choice of place than the hospital. On the other hand, death rate in the PICU was with 42% higher than the proportion of NICU deaths in our study. Again, some of the patients dying in a PICU are preterm neonates but a substantial proportion of patients from the other diagnostic groups are represented as well. We were especially surprised and concerned by the 36% of children from the neurology group.

Dying outside an ICU has been associated with the receipt of PPC. A retrospective study compared demographic and clinical characteristics associated with receipt of inpatient PPC using routine data from 24'342 children who died between 2001 and 2011. Data was extracted from a paediatric health database generated by more than 40 hospitals across the US. It was found that overall, children who received PPC were less likely to die in an ICU [40]. Changes in place of death were also observed in the course of time. Another retrospective study from a US tertiary level paediatric hospital compared data from a cohort of 102 children with cancer who died between 1990 and 1997 with a more recent cohort of 119 children who died between 1997 and 2004. Significantly fewer children from the more recent cohort died in an ICU [41]. More data from the US support this shifting place of death among children with a life-limiting CCC over time. In a big retrospective nationwide case series drawn from the National Center for Health Statistics' Multiple Cause of Death Files spanning from 1989 to 2003, it was observed that the odds of dying at home increased significantly each year [42].

It must be noted that paediatric death in an ICU does not imply poor quality of care but rather indicates a lack of choices presented to the families. Therefore an important mechanism explaining those shifting places of death for children with life-limiting CCCs can be attributed to elements of advanced care planning.

7.5.2. Advanced care planning

Advanced care planning, i.e. planning for care at EOL is a core element of PPC and has been described and recommended by the 2003 Institute of Medicine report "When children die" [18]. The purpose of care planning is to offer choices to families and to set goals applying a family-centred and integrated model of care [18, 43]. Advanced care planning has been linked to earlier discussions about the child's EOL including preferred location of care and death, and earlier documentation of do not resuscitate (DNR) orders before death [41, 44]. Advanced care planning can only succeed when crucial goals and decisions are well documented and available to the range of providers that are involved in an individual's child EOL care. In a recent chart review including a set of 114 notes relating to a cohort of 48 children from the UK who died between 2008 and 2010 within a defined geographical area, it was found that current documented EOL care planning varied between services [44]. When plans were documented, the information was often unstructured and in narrative form buried in the patient's healthcare record. Even though certain templates for DNR orders or child and family wishes existed, their existence alone did not ensure its adoption into practice [44].

The involvement of different healthcare providers creates not only challenges regarding the planning and documentation of care but the provision of continuous and coordinated care in general, a domain identified as being important to families of a dying child [45].

7.5.3. Continuity and coordination of care

Parental experiences as well as satisfaction with care related to continuity and coordination of care were rated lowest among all six quality domains in the PELICAN II study. Many of the listed negative experiences related to relational aspects of continuity of care such as frequent changes of physicians and nurses. Continuity of care has been conceptualised and three types were identified: (1) Informational continuity; (2) Relational continuity; and (3) Management continuity [46]. Relational continuity refers to "the importance of knowledge of the patient as a person; an ongoing relationship between patients and providers is the undergirding that connects care over time and bridges discontinuous events."[46, p. i].

Since so many patients spend substantial time during their EOL phase in an ICU, relational continuity in this care setting seems particular relevant. The delivery of continuity of nursing care in the PICU from the perspective of both parents and nurses was recently explored in a qualitative study from the USA [47]. The authors concluded that the family's desire for continuity of care often competed with how care is structured in the PICU environment. As PICUs are mostly part of a teaching hospital, the learning needs of the staff members often took precedence over relationship development and skill development was of primary importance. The authors however agreed that the origins of deficits in relational continuity was likely multidimensional and aspects like staffing fluctuations and fear of emotional entanglement added to the discrepancy of parental needs and the ability of nurses to meet those needs [47].

If and to what extent that study's result are transferable to experiences of the parents participating in PELICAN II is arguable. For a deeper insight and understanding of the PELICAN II results related to continuity of care, the results of the qualitative study arm will be connected and help explaining this quantitative result (not part of this dissertation). As for care coordination, this aspect becomes most relevant when several providers are involved in caring for the child and his/her family and especially, when outpatients care, e.g. home-based care is available.

7.5.4. Home-based PPC

Derived from the high proportion of hospital deaths in the PELICAN study, only a few children (17%) died at home. A higher proportion of 48% of the patient spent at least one day at home during the last four weeks of life, and half of those, received home-based nursing care. Only half of the children who spent some time at home during their EOL phase receiving professional home care seem to be a low number. Based on these quantitative results we could not infer whether this circumstance reflected the family's choice or was rather a result of limited access. Absence or inadequate home care services was identified as one of several barriers for EOL care at home in the PELICAN HOME sub-study based on single interviews with parents who cared for their dying child at home (unpublished result). Paediatric community care organizations providing paediatric care are well established in Switzerland. They are however massively challenged by funding structures and processes resulting in lacking compensation, and are often limited in their geographical and daytime range of action. These limitations collide with the major need of families for individually tailored support and flexibility in care during the EOL phase of their child. In a recent integrative review aiming at identifying the key elements of optimal models of paediatric palliative care, access to a specialised paediatric team was considered crucial by parents, as was specialist support around the clock [48]. A positive impact on the QOL of children and their parents and on the burden of home care for families of severely ill children has been demonstrated by the provision of paediatric palliative home care by a specialised team [49]. The team under inquiry was established at a centre for PPC in Germany and consisted of trained specialists in PPC such as paediatricians, nurses, a social worker and chaplain. A 24/7 on-call service, psychosocial support and coordination of care were the team's main tasks besides the provision of medical and nursing care [49].

Our study results indicate that the family's need for access to specialised EOL home care services is not met. Home is the preferred place to be for most children, but without appropriate support at home, parents might opt for hospital care instead.

7.6. Strengths and limitations of methods used

This dissertation was part of the nationwide PELICAN study, an extensive and comprehensive needs assessment of paediatric EOL care needs in Switzerland. Some of the PELICAN study results are influenced by the context, in which care is provided, e.g. health care system, care models. This limits generalisability internationally. However, the diagnostic group categorisation and group comparisons generated knowledge that is novel and valuable for healthcare providers offering services within the entire field of paediatrics, as is often the case for specialised PPC teams. Strengths and weaknesses of the single studies have been described in chapters 4 – 6. Apart from that, some topics warrant more emphasis and will be further discussed in the next paragraphs.

7.6.1. Recruitment and study participation

The major strength of the PELICAN study was the inclusion of all children who died due to a life-limiting CCC or prematurity, covering all major conditions responsible for childhood death. Additionally, the study was conducted in the entire country including patients and families from all main Swiss language regions. Thus, the results provide urgently needed information that is representative for Switzerland. Recruitment was conducted by all Swiss children's hospitals and paediatric community care organisations. In Switzerland, treatment of children with a life-limiting CCC is organised by hospital-based paediatric specialists. Hence we are confident that all children matching the PELICAN study's inclusion criteria were identified and that the sample can be considered population-based. However, it is known that clinicians, by knowing their patients and their families, sometimes decide that research participation might not be reasonable for certain families. They then act as gatekeeper and impede recruitment of those families [50]. We are aware of only one example, where this was the case in the PELICAN study. Another potential source of bias lies in study participation, and this was a more realistic threat in the PELICAN study. Study results might have been biased in that only parents with favourable experiences may have been motivated to participate. In accordance with ethical guidelines, study participation was entirely voluntary and we did not ask for reasons of non-participation.

The requirement of being proficient in German, French or Italian excluded some migrant residents representing cultural minorities in Switzerland. This is of concern as notable differences in location of death across racial and ethnic groups in the US have been described in children with life-limiting CCCs in general [42], and recently again in children with cancer [51]. This raises questions regarding equity of paediatric EOL care which need to be addressed in future evaluation of care and research.

7.6.2. Questionnaire development and testing

For the development and testing of the PaPEQu, rigour procedures were applied following existing recommendations. The questionnaire will need revisions for future use, mainly due to the conceptual weaknesses related to the assessment of parental needs. Nevertheless, the PaPEQu shows promise as a tool to retrospectively assess the parental perspective of EOL care their child and family received.

The PaPEQU was developed in German and content validity was established before translation into French and Italian. In the following pilot test including parents that lost their child in 2010, the sample for the French version was small and in the Italian-speaking part of Switzerland no childhood death was registered for that year. Although we emphasized the harmonisation step during the translation process ensuring that there were no translation discrepancies that arose between the different languages, content validity of the French and Italian PaPEQu versions is not fully established and the instrument should not be used to explore differences in paediatric EOL care between the three main Swiss language regions.

7.7. Implications for policy, practice and future research

Fortunately rare, childhood deaths due to illness or prematurity are still a reality and part of our health care system. Advances in medicine and technology, lacking awareness in society and a social perception of unnaturalness when a child dies all contribute to the medicalized deaths we see in paediatrics today. The contribution of lacking PPC models in Switzerland, incorporating advanced care planning, is however, equally important. The PELICAN study provides urgently needed country-specific basic knowledge which allows for planning and implementing the next steps in improving paediatric EOL care in Switzerland.

7.7.1. Policy

The gaps related to paediatric PC have been recognised in the" National Strategy for Palliative Care 2013-2015" [52]. In the framework of the Federal Office of Public Health that guides implementation of PC in Switzerland, PPC is categorized on the level of specialised care [53]. This means that in order to meet the complex health care needs of those patients and their families, a specialised HCT should be available. Specialised teams are formed by specialised professionals, and specialised professionals are formed by appropriate education and training. With the promotion of PC as medical sub-specialty and the creation of professorships at universities, palliative medicine started to be incorporated into the formal medical training. Similar offerings for nurses however are still vastly missing. The incorporation of PC in non-university health and social professions is part of the action plans within the sub-project "Training" of the "National Strategy for Palliative Care 2013-2015" [52]. Also, in accordance with the multiprofessional approach of PC, efforts to create shared training for different professional groups seem to be the way to go. On how this will be achieved however, remains to be seen.

Another development on the national and cantonal level(s) of Switzerland involve tarification of PC with the goal to create a performance-related structure that is uniformly applicable throughout Switzerland [52]. This is an important and needed endeavour as specific PC interventions are not represented well enough in existing codes. As always in these matters, professionals involved are challenged to ensure that paediatric specificities are represented as well, which was a problem in the past and stands out to be an issue in PC as well.

7.7.2. Practice

Elements of optimal PPC for children and young people have been described based on an integrative review including empirical studies that reported primary data evaluating models of PPC from the perspectives of children or parents [48]. The elements identified as most important were "access to tailored support including flexibility in location of care, psychosocial care, 24h specialist support, respite care and support for siblings" [48, p. 430]. A consultative model of PPC, comprising a multiprofessional team of PPC specialists that provide specialist support and advice to the child's primary care team, might be well suited to address those identified elements. Concepts to such an approach exist and are implemented in only three children's hospitals (only two among the five Swiss university hospitals). Nevertheless, together with the PELICAN study results, those examples can serve as a good starting point in promoting and supporting further initiatives on the of hospital level and possibly on a regional or national level.

Flexibility in location of care and 24h specialist support have been identified as important elements of PPC and our study results raised important questions in terms of home-based care in Switzerland. The scope and range in which hospital-based PPC teams can operate need to be extended in order for them to reach patients at home or to collaborate closer with community-based nursing care and possibly general practitioners. The model from Germany described and evaluated by Groh et al. [49] might serve as another example to look at, as the coordination of professional assistance in cooperation with the local health care professionals and a 24/7 on-call service were described as main tasks of that multiprofessional team. Additionally, efforts needs to be put into the support related to financial issues. Refund processes of insurances work very slowly and put some families in situations, where financial shortcomings decide on location of care.

So what can be recommended to the individual health care provider who is inherently motivated to do good and support dying children and their families to the best of their knowledge and skills, but within the structural and management limits sometimes present in their institutions? Not an easy question to answer. I would tell them to stay true to themselves and be open and honest with the family to build up this trustworthy relationship that is so important. And always keeping in the back of their minds, that only one negative interpersonal event between the child or the parent and the health care professional is needed for potential lasting emotional distress in parents who lost a child.

7.7.3. Research

To only recommend that hospitals should develop and implement a presumed state of the art PPC programme seems too easy in the light of the current evidence base in terms of effectiveness. One result of the abovementioned review describing elements of optimal PPC was, that definitive information on effectiveness could not be provided due to the lack of high quality data [48]. In a performance-oriented health care system where financial resources are always in short supply and medical successes are cherished, for a hospital to decide to invest in PPC seems to be a tough call. A scenario in which solid outcome measures put the child and his/her family at the centre of care and demonstrate what meaningful differences PC can make might influence how funding is allocated. The call for introducing outcome measurement into practice has been issued by the task force on outcome measurement of the European Association for Palliative Care (EAPC) in their White Paper publication [54]. Concerning PPC it was mentioned that due to age- and cognitive function-related specifics in paediatrics, outcome measurement is particularly challenging and should be extended to parents and siblings [54]. Common outcome measures applied in adult PC include multidimensional tools to assess symptoms, distress and functioning indifferent areas. Although systematic pain assessment is well established in paediatrics, valid multidimensional tools for palliative care in children could not been identified by the EAPC task force [54].

Due to the particular situation in paediatric palliative and EOL care, where a child passes away early and leaves behind parents and siblings, it is my strong opinion that evaluation of PPC and EOL care has to go beyond measuring outcomes on the patient level. "Patient"-reported outcome measurement has to be extended and applied on the family level. However, consensus on meaningful outcomes on the family level has not yet been found. Self-reported QOL of parents and possibly siblings seems relevant and has been used before to evaluate PPC services [49]. However, I believe the mechanisms determining QOL are not yet well understood and should further be explored. This holds also true for satisfaction with care, considering the conceptual deficiencies described in paragraph 7.2.1. Themes of needs most important to parents are well defined and can be compiled to build a construct of good quality EOL care. The contribution and therefore importance, of each of the various domains to the construct however, is not yet clear and should further be tested. Further exploratory research to better define and establish measurement constructs should eventually evolve into intervention and the drawing of causal conclusions.

Following the multiprofessional approach in PC practice, I strongly support interprofessionality in research as well. The perspectives of e.g. physicians, adds to the relevance of questions asked. Related to EOL care outcomes, a special benefit could lie in the collaboration with psychologists. Bereavement research, which is occupied by the discipline of psychology, should be part of EOL care evaluation as bereavement support is an integral part of PPC. Considering that bereavement might be influenced by experiences made during the EOL of the child [1], it would be interesting to further explore factors influence that some families do better than others.

7.8. Conclusions

PPC has emerged as a medical subspecialty over the last decade. I builds on the principles of respect and dignity, focuses on ensuring the best possible QOL of life for the child and his/her family by extending beyond the physical domain into psychosocial and spiritual wellbeing [55]. To eventually grow out its infancy, PPC practitioners are challenged by demonstrating effectiveness of their services in performance-oriented healthcare systems with scarce resources.

The PELICAN study helped define the Swiss population in regard to the characteristics of children dying in various diagnostic groups, their circumstances of death and the experiences of their parents during the child's EOL care. This so far missing basis of data is needed to plan and implement a needs-driven and evidence-based model of PPC for Switzerland.

The inclusion of all relevant diagnoses responsible for illness-related childhood death and preamaturity allowed comparisons between four distinct diagnostic groups. This approach provided novel information related to circumstances of death and symptoms, and related to the parental perspecitve of their child's EOL care, which is valualbe to health care providers involved in paediatric EOL care worldwide.

7.9. References

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29. Streuli, J. and Bergsträsser, E., *"Shared Decision-Making" in der Kinder- und Jugendmedizin.* Paediatrica, 2015. **26**(4): p. 12-15.
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55. Liben, S., Papadatou, D. and Wolfe, J., *Paediatric palliative care: challenges and emerging ideas.* Lancet, 2008. **371**(9615): p. 852-64.

Curriculum Vitae

Karin Zimmermann

Date of Birth	December 23, 1969 in Bern
Civil Status	unmarried
Nationality	Swiss, from Wattenwil

Business Address

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Education

2012-2016	PhD in Nursing Science, Institute of Nursing Science, University of Basel
2008-2011	Master of Science Nursing, Institute of Nursing Science, University of Basel
2008	Online Course NIH “Human Participant Protections: Education for Research Teams”.
2006-2008	Bachelor of Science Nursing (BScN), WE’G Hochschule Gesundheit, Aarau, Switzerland
2006	Course in Good Clinical Practice, Inselspital, Bern, Switzerland
1997–1999	School of Advanced Nursing Level 1 Oncology, St. Gallen, Switzerland
1989–1992	School of Pediatric and Neonatal Nursing, Inselspital, Bern, Switzerland
1986–1988	Diplommittelschule Marzili, Bern, Switzerland
1981–1986	Secondary School Spiegel, Spiegel, Switzerland
1977–1981	Primary School Buchsee, Koeniz, Switzerland

Clinical Employment

05/2016 to date	University Children’s Hospital Zürich, Eleonorenstiftung, Paediatric Palliative Team, Scientific Collaborator
2014-2016	Inselspital Bern University Hospital, Directorate of Nursing/Nursing Research Unit, Scientific Collaborator
2003-2014	Inselspital Bern University Hospital, Department of Pediatrics Hematology/Oncology, Pediatric Advanced Nurse Level 1 BScN, 2006 – 2008 Clinical Research Associate, from 2011 Clinical Nurse Specialist MScN
2000–2003	PC-Joy Bern, Branch Manager
1994–2000	Inselspital Bern University Hospital, Department of Pediatrics, Pediatric Hematology/Oncology, Pediatric Nurse from 1999 Advanced Nurse Level 1 Oncology
1992–1994	Inselspital Bern University Hospital, Department of Pediatrics, Neonatology, Pediatric Nurse

Academic Employment

04/2016 to date	Institute of Nursing Science, University of Basel, Lecturer and scientific collaborator
2012-2016	Institute of Nursing Science, University of Basel, Research Assistant

Research Grants

CHF 9'500.-	**Zimmermann, K.,** Cignacco E., Bergsträsser, E. (2016). **P**aediatric **E**nd-of-**LI**fe **CA**re **N**eeds in Switzerland (**PELICAN** study, 2012-2015). **Parental Perspectives on their child's end-of-life care:** adding insights and richness to primarily quantitative results by mixing in qualitative results – Extension stipend award supported by the PhD Program Health Sciences (PPHS).
CHF 189'000.-	Bergstraesser E., Cignacco E., Von der Weid N., **Zimmermann K.**, Paediatric End-of-LIfe CAre Needs in Switzerland (PELICAN Study) (2012-2015) – Unterstützt durch Krebsforschung/Krebsliga Schweiz, Bern
CHF 100'000.-	Cignacco E., **Zimmermann K.**, Bergstraesser E. Paediatric End-of-LIfe CAre Needs in Switzerland (PELICAN Study) (2012-2015) – Unterstützt durch Stiftung für Pflegewissenschaft Schweiz, Basel
CHF 60'000.-	Bergstraesser E., **Zimmermann K.**, Cignacco E., Paediatric End-of-LIfe CAre Needs in Switzerland (PELICAN Study) (2012-2015) – Unterstützt durch Bundesamt für Gesundheit, Bern
CHF 25'000.-	**Zimmermann, K.** (2012). Paediatric End-of-LIfe CAre Needs in Switzerland (PELICAN) – Start stipend award supported by the PhD Program in Health Sciences.

Publications – Original work peer review

Zimmermann, K., Bergstraesser, E., Engberg, S., Ramelet, A. S., Marfurt-Russenberger, K., Von der Weid, N., Grandjean, C., Fahrni-Nater, P., Cignacco, E., & Consortium, Pelican. (2016). When parents face the death of their child: a nationwide cross-sectional survey of parental perspectives on their child's end-of life care. BMC Palliative Care, 15(1), 30. doi: 10.1186/s12904-016-0098-3

Zimmermann, K., Cignacco, E., Eskola, K., Engberg, S., Ramelet, A. S., Von der Weid, N., & Bergstraesser, E. (2015). Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu) - an instrument to assess parental experiences and needs during their child's end-of-life care. Journal of Advanced Nursing, 71(12), 3006-3017. doi: 10.1111/jan.12741

Graf, E., Cignacco, E., **Zimmermann, K.**, & Zuniga, F. (2015). Affective Organizational Commitment in Swiss Nursing Homes: A Cross-Sectional Study. Gerontologist. doi: 10.1093/geront/gnv053

Gianinazzi, M. E., Rueegg, C. S., **Zimmerman, K.**, Kuehni, C. E., Michel, G., & Swiss Paediatric Oncology, G. (2015). Intra-Rater and Inter-Rater Reliability of a Medical Record Abstraction Study on Transition of Care after Childhood Cancer. PloS One, 10(5), e0124290. doi: 10.1371/journal.pone.0124290

Eskola, K., Bergstraesser, E., **Zimmermann, K.**, & Cignacco, E. (2015). Paediatric end-of-life care in the home care setting (PELICAN HOME)--a mixed methods study protocol. Journal of Advanced Nursing, 71(1), 204-213. doi: 10.1111/jan.12463

Bergstraesser, E., **Zimmermann, K.**, Eskola, K., Luck, P., Ramelet, A. S., & Cignacco, E. (2015). Paediatric end-of-life care needs in Switzerland: current practices, and perspectives from parents and professionals. A study protocol. Journal of Advanced Nursing, 71(8), 1940-1947. doi: 10.1111/jan.12650

Zimmermann, K., Ammann, R. A., Kuehni, C. E., De Geest, S., & Cignacco, E. (2012). Malnutrition in pediatric patients with cancer at diagnosis and troughout therapy: a multicenter cohort study. Pediatric Blood & Cancer, 2013. **60**(4): p. 642-9.

Michel, G., Kuehni, C. E., Rebholz, C. E., **Zimmermann, K.**, Eiser, C., Rueegg, C. S., et al. Can health beliefs help in explaining attendance to follow-up care? The Swiss childhood cancer survivor study. Psychooncology, 20(10), 1034-1043. doi: 10.1002/pon.1823

Zimmermann, K. (2008). [Constipation management in the pediatric oncology patient]. Pharm Unserer Zeit, 37(2), 156-160. doi: 10.1002/pauz.200700259

Publication – Non-Databased, not peer review

Zimmermann, K. (2015). [End filtration in infusion therapy]. Krankenpflege. Soins Infirmiers, 108(11), 34-35.

Zimmermann, K., Eskola, K., & Dorsaz, A. (2014). [When children die]. Krankenpflege. Soins Infirmiers, 107(10), 78-79.

Zimmermann, K., Eskola, K., & Dorsaz, A. (2014). [End of life care in pediatrics: when children die]. Krankenpflege. Soins Infirmiers, 107(3), 38-39.

Zimmermann, K. (2014). Research Flash: Medikamentenfehler im häuslichen Setting bei Kinder. Onkologiepflege / Soins en oncologie / Cure oncologiche, 2014(1), 18-19.

Frei, M., & **Zimmermann, K.** (2014). Komplementärmedizin und Kinderonkologie: Umfrage bei Fachpersonen in pädiatrischer Onkologiepflege. Onkologiepflege / Soins en oncologie / Cure oncologiche, 2014(2).

Zimmermann, K. (2013). Beste Masterarbeit 2011/2012: Malnutrition bei pädiatrisch-onkologischen Patienten. Onkologiepflege: Fachzeitschrift der Onkologiepflege Schweiz, 2013(3).

Zimmermann, K. (2013). Kinder-Palliative Zentrum. Onkologiepflege: Fachzeitschrift der Onkologiepflege Schweiz, 2013(2).

Zimmermann, K. (2009). Obstipationsmanagement bei pädiatrisch-onkologischen Patienten - Resultate der Untersuchung 2008 - Erkenntnisse in Bezug auf das Assessment. Onkologiepflege(2)

Publications – Book Chapters

Cignacco, E., Bergsträsser, E., **Zimmermann, K.**, Eskola, K., Luck, P., Marfurt-Russenberger, K., Dorsaz, A., Fahrni-Nater, P., & Ramelet, A.S. (2015). PELICAN – Die erste nationale Studie über die Betreuung von Kindern am Lebensende. In Stiftung für Pflegewissenschaft (Ed.), 10 Jahre Stiftung Pflegewissenschaft Schweiz (pp. 101-111). Basel, Schweiz: Stiftung für Pflegewissenschaft.

Published Abstracts

Zimmermann, K., Bergstraesser, E., & group, on behalf of the PELICAN study. (2016). *Paediatric End-of-LIfe CAre Needs in Switzerland - PELICAN (2012-2015): Description of Current Practices (PELICAN I) and Assessment of Parental Perspectives (PELICAN II quantitative)*. Abstract published in Palliative Medicine, 30(6). doi: 10.1177/0269216316646056.

Zimmermann, K., Ammann, R.A., Kuehni, C.E., De Geest, S., & Cignacco, E. (2012). *Prevalence of malnutrition in pediatric patients with cancer: A multicenter cohort study*. Abstract published in Pediatric Blood & Cancer, 59(6), doi: 10.1002/pbc.24295.

Master Thesis

Zimmermann, K. (2011). Malnutrition in pediatric oncology patients at diagnosis and throughout therapy: a multicenter cohort study. Master of Science Nursing, University of Basel, Basel.

Bachelor Thesis

Zimmermann, K. (2008). Obstipationsmanagement auf pädiatrisch-onkologischen Abteilungen der Schweiz. Bachelor of Science Nursing, WE'G Hochschule Gesundheit, Aarau.

Presentations – International

Zimmermann, K., Bergstraesser, E., & group, on behalf of the PELICAN study. (2016). *Paediatric End-of-LIfe CAre Needs in Switzerland - PELICAN (2012-2015): Description of Current Practices (PELICAN I) and Assessment of Parental Perspectives (PELICAN II quantitative)*. Poster presented at the 9th World Research Congress of the European Association for Palliative Care, (Ireland), May 09-11.

Zimmermann, K. (2013). Malnutrition in der pädiatrischen Onkologie. Oral presentation at Jahrestagung der Deutschen, Österreichischen und Schweizerischen Gesellschaft für Hämatologie und Onkologie, (Austria), October 19-20.

Zimmermann, K. (2012). Prevalence of malnutrition in pediatric patients with cancer: a multicenter cohort study. Oral presentation at the 44th congress of the International Society for Paediatric Oncology (United Kingdom), October 5-8.

Zimmermann, K., Kellerhals, S., Fliedner, M. (2012). Care specific aspects of the Swiss stem cell transplantation centers. Oral presentation at the EMBT 2012 congress (Switzerland), April 3.

Zimmermann, K. (2012). Prevalence of malnutrition in pediatric-oncology. Oral presentation at the international conference "Challenges in Pediatric Care: Innovations through Advanced Nursing Practice" (Switzerland), January 13.

Zimmermann, K. (2011). Obstipationsmanagement bei pädiatrisch-onkologischen Patienten: Entwicklung einer klinischen Leitlinie. Oral presentation at Fachtagung für die Hämatologisch – Onkologische Kinderkrankenpflege (Germany), September 30-October 1.

Presentations – National

Bergsträsser, E., **Zimmermann, K.** (2016). Results of the PELICAN Study (Paediatric End-of-LIfe CAre Needs in Switzerland). Oral presentation at the SPOG Scientific Meeting (Bern), June 03.

Zimmermann, K., Bergsträsser, E., Brazzola, P.L. (2016). PELICAN study: Framework Concept for Palliative Care in Switzerland – Current Situation. Invited lecture at the Annual meeting of the Swiss Society of Neuropediatrics (Lugano), May 18.

Bergsträsser, E., **Zimmermann, K.** (2015). PELICAN – Pädiatrische End-of-Life Care Bedürfnisse in der Schweiz. Erste Resultate. Invited lecture at Nationale Palliative Care Tage, (Bern), Switzerland.

Zimmermann, K.; Cignacco, E.; Eskola, K.; Luck, P.; Bergsträsser, E. (2014) PELICAN – Paediatric End-of-LIfe CAre Needs in Switzerland. Oral presentation at VFP Fachtagung "Pflege bewegt" (Bern), April 01

Zimmermann, K.; Cignacco, E.; Eskola, K.; Luck, P.; Bergsträsser, E. (2013) PELICAN – Paediatric End-of-LIfe CAre Needs in Switzerland. Poster submitted to “Nationaler Forschungstag Palliative Care” (Bern), November 26.

Zimmermann, K. (2012) PELICAN – Paediatric End-of-LIfe CAre Needs in Switzerland. Oral presentation at Symposium Kispi Zurich: Palliative Care bei Kindern (Zurich), December 06.

Zimmermann, K., Chettata, S. (2012). Malnutrition: Prävalenz, Assessment, Support. Oral presentation at Fachtagung für Fortgeschrittene in pädiatrischer Onkologiepflege (Bern), November 09 and May 30 2013.

Zimmermann, K., Chettata, S. (2012). Ernährung und Verdauung. Workshop at Pflegefachtagung Pädiatrie 2012 (Luzern), September 27.

Zimmermann, K. (2012) Malnutrition in der pädiatrischen Onkologie. Oral presentation at SBK Kongress 2012 (Interlaken), May 10.

Zimmermann, K. (2012). Malnutrition in pediatric oncology patients at diagnosis and throughout therapy: a multicenter cohort study. Oral presentation at the SPOG Scientific Meeting (Lugano), January 27.

Zimmermann, K. (2011). Nationale klinische Leitlinie zum Obstipationsmanagement bei pädiatrisch-onkologischen Patienten. Oral presentation at the SPOG Scientific Meeting (Lugano), February 5.

Zimmermann, K. (2010). Klinische Leitlinie Obstipation. Oral presentation at Fachtagung für Fortgeschrittene in Pädiatrischer Onkologiepflege (Luzern), November 11.

Zimmermann, K. (2010). Malnutrition in pediatric oncology patients at diagnosis and during therapy: a retrospective multicenter study. Oral presentation at the SPOG Scientific Meeting (Lugano), January 29.

Zimmermann, K., Julmy, F., Leibundgut, K. (2010). Safe administration of complex chemotherapy regimens via intravenous in-line filters. Oral presentation at the SPOG Scientific Meeting (Lugano), January 29.

Awards

Zimmermann, K.; Cignacco, E.; Eskola, K.; Luck, P.; Bergsträsser, E. (2013) PELICAN – Paediatric End-of-LIfe CAre Needs in Switzerland. Poster submitted to “Nationaler Forschungstag Palliative Care” (Bern), November 26, second poster prize.

Zimmermann, K. Best master thesis 2011 „Malnutrition in Pediatric Oncology Patients at Diagnosis and throughout Therapy: A Multicenter Cohort Study“, The Nursing Science Foundation Switzerland.

Memberships

2014 to date	Member of the board of the Swiss Cancer League
	Associate Member Children’s Oncology Group (COG): Nurses Group und Clinical Research Associates Group.
	Member of the International Society of Pediatric Oncology (SIOP)
	Member of Onkologiepflege Schweiz

Review Activities

Scientific Journals

2015 to date	Reviewer of the “Journal of Advanced Nursing”
2015 to date	Reviewer of the “International Journal of Nursing Studies”

Organization of Workshops & Scientific Conferences

2016-2017	National PELICAN conference, February 2017 (Member of the organizing committee).
2015-2016	National conference Swiss Pediatric Nursing, (President of the organizing committee).
2014	Visionstag Pädiatrische Pflege und Pflegeforschung Switzerland, April 09, (Member of the organizing committee).
2010-2011	International conference “Challenges in Pediatric Care: Innovations through Advanced Nursing Practice”, January 13 2012, (Member of the organizing committee).
2008 - 2014	Nationale Fachtagung für Fortgeschrittene in pädiatrischer Onkologiepflege, yearly, (Member of he organizing committee).

Teaching Assignments

University of Basel, Faculty of Medicine, Institute of Nursing Science

2016	Research Methodology/Methods I and II
2014	ANP Course
2013, 2014	Master Seminar quantitativ
2013, 2014, 2015	Seminar “Proposal Writing” quantitativ
2012, 2013, 2014	Scientific Writing Course
2012	Coaching “Clinical Leadership”

Onkologiepflege Schweiz, Basiskurs der Fachgruppe Pädiatrische Onkologiepflege Schweiz (POPS)

Yearly-2015	Constipation

Last Update: Jaberg, September 2016