Specialised **P**aediatric P**A**lliativ**E** Ca**R**e: **A**ssessing **E**conomic outcomes in a multi-site context of various care settings (**SP**h**AERA**-**E**)

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List of Abbreviations

CHF	Swiss francs
CHOP	Swiss classification of operations
CI	Confidence interval
Coef.	Coefficient
CPI	Consumer price index
DCGM	DISABKIDS chronic generic measure
DRG	Diagnosis related group
FHI	Family hardiness index
FSOC	Family sense of coherence
FTE	Full-time equivalent
HCP	Healthcare professional
IQR	Inter quartile range
KVG	Bundesgesetz über die Krankenversicherung (Federal Health Insurance Act)
KLV	Krankenpflege-Leistungsverordnung (Patient Care Services Regulation)
LLC	Life-limiting condition
LoS	Length of stay
MSAS	Memorial symptom assessment scale
OKP	Obligatorische Krankenpflegeversicherung (Health Care Benefits Ordinance)
OOP	Out-of-pocket
PaPaS-Scale	Paediatric palliative screening scale
PC	Palliative care
PICU	Paediatric intensive care unit
PPC	Paediatric palliative care

PRISMA	Preferred reporting items for systematic reviews and meta-analyses
ProQOL	Professional quality of life
QOL	Quality of life
QOLLTI-F	Quality of life in life threatening illness–family carer
READ	Ready materials; Extract data; Analyse data; Distil findings
Rng.	Range
SD	Standard deviation
SPhAERA	Specialised Paediatric PAlliativE CaRe: Assessing family, healthcare professionals and health system outcomes in a multi-site context of various care settings
SPPC	Specialised paediatric palliative care
TARMED	Tarif médical
TfSL	Together for Short Lives
TV	Television
USD	United States dollar
WHOQOL	World Health Organisation quality of life
WüTi	Würzburger Trauerinventar (Würzburg Grief Inventory)

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Summary

Over the last decades, the prevalence of life-limiting conditions (LLCs), conditions for which there is no reasonable hope of cure and where premature death is likely, in children has been increasing. As parenting a child with an LLC is an enormously challenging and demanding experience, parents of these children are at an increased risk of profound caregiver burden. Besides physical, psychological, emotional and social burdens, this also includes financial burden from out-of-pocket medical and non-medical expenses and employment-related income loss. However, there is an absence of literature on how illness-related events, such as a child's hospitalisation or even death, contribute to these costs. Moreover, inconsistencies in the outcome measures and methodological approaches used to quantify financial burden limit the interpretation and comparability of research findings, making it inherently difficult to draw conclusions about the magnitude of the financial burden experienced by affected families.

Nevertheless, robust evidence on the sources and determinants as well as the true magnitude of families' financial burden is crucial for enabling care and support interventions, such as specialised paediatric palliative care (SPPC), to effectively address families' financial support needs. SPPC is advocated as a standard of care for children with LLCs and their families. However, despite the acknowledged benefits of SPPC, funding constraints have emerged in Switzerland, which likely limit its accessibility and provision to families in need.

Therefore, this dissertation investigated existing measures of financial burden in families of children with LLCs, explored hospitalisation- and death-related financial and employment implications for these families and examined current funding structures, obstacles and priorities regarding the funding of SPPC programmes in Switzerland.

In conducting this dissertation, the published literature investigating financial costs incurred by families of children with LLCs was reviewed, economic data from the larger 'Specialised Paediatric PAlliativE CaRe: Assessing family, healthcare professionals and health system outcomes in a multi-site context of various care settings (SPhAERA)' study was analysed, and an expert panel on the provision and funding of SPPC in Switzerland was consulted.

Overall, this dissertation encompasses five chapters:

The first chapter of this dissertation provides an overview of the definition and prevalence of LLCs in children and embeds financial burden within the wider context of parental caregiving, including the multiple caregiver burdens experienced by parents. This sets the stage for an

introduction of the conceptual framework guiding this research. This, in turn, is followed by a description of SPPC and a brief presentation of the SPhAERA study. A presentation of the rationale and aims concludes **Chapter 1**.

The scoping review presented in **Chapter 2** provides an overview of cost indicators and outcome measures used to measure financial burden in families of children with LLCs. The retrieved cost indicators and outcome measures were categorised into three broad groups: direct costs, that is, out-of-pocket medical and non-medical expenses; indirect costs and financial support. The review suggests that not all key components of families' financial burden were consistently measured across reviewed studies. To facilitate future investigations of financial burden, a framework for comprehensively measuring families' out-of-pocket medical and non-medical expenses was developed, and clear recommendations for assessing income loss, opportunity costs, and financial support were provided.

Chapter 3 reports the results of a cohort study that investigated hospitalisation- and death-related financial and employment implications experienced by parents of children with LLCs. Over care and bereavement follow-up assessments of 330 and 300 days, respectively; a broad range of out-of-pocket non-medical expenses and employment-related outcomes were analysed using descriptive statistics and fixed-effects regressions. The analysis included 59 mothers and 51 fathers of 61 children. The results showed that families incur higher travel and accommodation expenses during hospitalisation than during non-hospitalised periods. In addition, during the first 120 days of bereavement, more than one-fifth of grieving parents increased their work commitments.

In the research presented in **Chapter 4**, a four-step conceptualisation process, including a document analysis was used to conceptualise the funding of hospital-based consultative SPPC programmes in Switzerland. The results revealed that current funding structures are complex and fragmented, combining funding from public, private and charitable sources. In addition, in consultation with an expert panel, a wide range of obstacles to and priorities for funding these programmes sustainably were identified. Overall, 21 experts participated in the modified Delphi study and identified 23 obstacles and 29 priorities. The highest level of consensus (>90%) was achieved for three priorities: the development of financing solutions to ensure long-term funding of SPPC programmes; the provision of funding and support for integrated palliative care; and the sufficient reimbursement of inpatient service costs in the context of high-deficit palliative care patients.

Chapter 5 provides a synthesis and discussion of the findings of this dissertation. The chapter starts with a discussion of the sources and determinants of financial burden in families of children with LLCs. This is followed by a discussion of how SPPC programmes can detect and actively address families' financial burdens. In this regard, the chapter also discusses potential implications of funding shortfalls on the provision of support aimed at addressing families' financial support needs. This in turn is followed by a brief discussion of contextual factors. Furthermore, the strengths and limitations of this research are examined while also presenting implications for future research, policy and practice.

In conclusion, SPPC programmes can help to address the financial support needs of families of children with LLCs. To enable these programmes to provide proactive support, it is essential that they are equipped with sufficient financial and personnel resources. In Switzerland, where SPPC programmes face major funding challenges, policy makers should establish an adequate financial and regulatory framework that facilitates the widespread implementation and provision of SPPC.

Chapter 1: Introduction

1.1 Introduction

In recent decades, progress in life-extending medical treatment and technology has led to a rising prevalence of life-limiting conditions (LLCs) in children.[1-3] Estimates for England indicate an increase in prevalence from 26.7 to 66.4 per 10'000 population (children 0–19 years) between 2001/2002 and 2017/2018.[2] Parenting a child with an LLC, that is, conditions for which there is no reasonable hope of cure and where premature death is likely,[4] is an extremely demanding and challenging experience.[5-7] Parents (including legal guardians) have to navigate the extensive care and support needs of their child, while coping with the emotional impact of their child's LLC at the same time.[8,9] As a consequence of the extensive caregiving demands and emotional distress, parents are at an elevated risk of severe caregiver burden,[6,7] frequently experiencing profound physical, psychological, emotional, and social adversities.[10-14] In addition, parents of children with LLCs face an increased risk of financial burden.[15] Evidence from paediatric oncology suggests that parents endure substantial out-of-pocket (OOP) medical and non-medical expenses and employment-related income losses.[16,17] However, longitudinal studies with multiple assessments that provide empirical evidence of how the costs endured by families of children with LLCs evolve over time are lacking. Yet, such evidence is pivotal for identifying and comprehensively addressing the unmet financial support needs of parents through effective interventions, for example, delivered by specialised paediatric palliative care (SPPC) teams. SPPC has been recommended as a standard of care for children with LLCs and their families.[18] In Switzerland, however, funding issues may impede the provision of SPPC. This may not only limit patients' and their families' access to medical and psychosocial care and support but also to needed financial support.

1.2 Life-limiting conditions in children

1.2.1 Definition

According to Together for Short Lives (TfSL), LLCs in children encompass four groups of conditions: (1) conditions for which curative treatment may be feasible but can fail, for example, cancer and irreversible organ failure; (2) conditions in which premature death is inevitable but long periods of intensive treatment are likely, for example, Duchenne muscular dystrophy; (3) progressive conditions without curative treatment options, for example, severe metabolic conditions; and (4) irreversible but nonprogressive conditions that cause severe disability, for

example, severe cerebral palsy.[4] As TfSL notes, the examples provided for each group are neither rigid nor exhaustive.[4] Over the course of illness, children may move between groups or belong to more than one group simultaneously.[4] In addition, efforts have been made to define LLCs and identify children with LLCs using the International Classification of Diseases.[1,19] For England their data indicates that the most common diagnoses of LLCs in children include congenital anomalies (31%), cancer (14%) and neurological conditions (12%), followed by blood disorders (10%) and respiratory conditions (9%).[1]

In the literature, the terms 'life-limiting' and 'life-threatening' conditions are often used inter-changeably, without clear distinction.[20] However, unlike 'life-limiting' conditions, which are chronic in nature, 'life-threatening' conditions refer to children at an acute risk of death, i.e., it refers to a specific moment in illness when a child faces an immediate threat to life.[20] TfSL defines 'life-threatening' conditions as '*those for which curative treatment may be feasible but may fail*'.[4,p.10] Throughout this dissertation, the term 'life-limiting' conditions as defined by TfSL, that is, the four groups of conditions presented above, is used.

1.2.2 Prevalence

Globally, only a few studies have attempted to estimate the prevalence of LLCs in children. Estimations from the United Kingdom,[1,2] Australia[3] and Germany[21] indicate that the number of children with LLCs has been rising in recent decades. In England an increase in prevalence from 26.7 to 66.4 per 10'000 population (children 0–19 years) between 2001/2002 and 2017/2018 has been estimated,[2] while for Queensland, Australia, estimates indicate an increase in prevalence from 35.2 to 43.2 per 10'000 (children 0–21 years) between 2011 and 2016.[3] Regarding Switzerland, a study on the prevalence of LLCs in children, following the approach of Fraser et al.[1,2] is currently underway at the Federal Office of Public Health. However, doing a preliminary extrapolation based on Fraser et al.'s[2] latest prevalence data and the Swiss population of 2019,[22] up to 11'200 children (0–19 years) live with an LLC in Switzerland.

While these estimations are based on hospital admissions data only, a more comprehensive estimation in Germany using data from 2019 insurance claims of both inpatient and outpatient care indicates that the prevalence of LLCs in children (0–19 years) could be as high as 262.2 per 10'000 population.[21] They noted that differences in prevalence estimations may be due to differences in measurement approaches, medical coding practices and differences in national population characteristics.[21] Nevertheless, due to advances in life-extending medical care and

technology leading to prolonged life-spans, the prevalence of LLCs in children is likely to further increase in the future.[2]

1.3 Parental caregiving

Caring for a child with an LLC is an extremely challenging experience for parents, which often comes with profound caregiver burden, including financial burden.[10-15] To gain a deeper understanding of the financial burden faced by parents, it is essential to acknowledge the caregiving context in which this burden arises. This includes recognising the location where parents provide care (i.e., place of care), as well as understanding the demanding roles and responsibilities that parents assume, which may vary based on the place of care. In addition, to gain a comprehensive understanding of the impact of financial burden, it is essential to consider it alongside other caregiver burdens faced by parents, for example, the physical and/or emotional burden.

1.3.1 Place of care

Depending on the disease's characteristics, children with LLCs must cope with multiple and complex symptoms, including pain, severe physical and functional limitations, technology dependency and medical frailty that affect every aspect of a child's life and the lives of their families.[23-26] Thereby, the complex care needs of children with LLCs extend beyond clinical treatment; they also include psychosocial and spiritual aspects and require holistic family-centred care provided in dedicated settings.[27,28] Care and support for children with LLCs is typically provided in the family's home, the hospital or in other (health-)care facilities, such as rehabilitation centres, residential facilities or hospices.[29] Regarding children's place of care, an analysis of Italian hospital discharge data showed that over 90% are discharged to their home.[30] In a more recent study conducted in Germany, this number was 77% for children receiving SPPC.[31]

These numbers align with parents' place of care preference, as supported by evidence indicating that most parents prefer to care for their child at home.[25,32-34] Parents were found to perceive caregiving in the family's home as less stressful because they felt more in control, had more privacy, could better maintain their daily activities and had greater opportunities for social engagement.[25,33] On the other side, parents expressing a preference for (health-)care facilities as

their child's primary place of care highlighted the value of receiving consistent, professional care and support.[25] However, due to the constrained availability of specialised facilities, such as long-term care facilities, parents' options to choose between care locations are limited in many countries.[35,36]

As children with LLCs often experience extended and recurrent hospitalisations, they spend a considerable amount of time in hospital.[26,30,37-41] The length of hospital stays tend to be particularly high in younger age groups (for non-oncological LLCs)[30] and towards the end-of-life.[37] Children with cancer were found to be hospitalised more frequently compared to children with non-oncological LLCs.[30] The reasons for hospitalisations of children with LLCs arise from various factors relating to illness management, the performance of diagnostic or therapeutic procedures and the occurrence of complications.[30]

As the following subchapter will outline, the place of care for a child with an LLC profoundly influences how parents comprehend and carry out their caregiving roles and responsibilities.[32] It shapes how parents navigate care, manage caregiving demands and engage in caregiving activities.[32]

1.3.2 Roles and responsibilities

While the hospitalisation of a child is a stressful event for parents, they usually desire and expect to be involved in their child's hospital care.[42] Parents want to actively engage in activities that provide them with a sense of familiarity and continuity of their usual caregiving routines at home, for example, feeding, comforting and providing emotional support.[42] In addition, parents were found to take on responsibilities for ensuring that treatments are on schedule, coordinating and keeping contact with healthcare professionals and staying informed about new treatment options.[42,43] However, to accommodate for one or both parents keeping a bedside presence at the hospital, families are required to adapt in their roles and responsibilities at home.[44,45] Child hospitalisations have been found to disrupt a family's daily routines, requiring them to reorganise household duties, modify work schedules and organise childcare for their other children.[44,45]

A child's transition from hospital to home can be complex and requires careful discharge planning, including close communication and coordination with families and community services.[46,47] Adequate parental preparation is crucial in supporting them to fulfil their role as their

ill child's caregiver in the family's home.[46,47] This includes educating parents about medication use and how to identify symptoms of illness, training them on how to use clinical equipment and preparing them to navigate emergency situations.[29,46,47]

Complex medical care at home places substantial responsibilities on parents.[48] Taking care of a child with an LLC has been described as '*managing an unexpected life*'[9,p.4] by parents, who must master multiple roles that exceed traditional parenting responsibilities.[49] These roles include, but are not limited to, medical caregiver, coordinator (e.g., appointment scheduling and managing appointments), educator (e.g., training other family members on using medical equipment), and financial and insurance navigator.[47,49] These roles entail a tremendous amount of demanding and time-consuming physical and mental work.[8,49,50] A qualitative interview study with parents of children with LLCs receiving palliative care at home identified several caregiving tasks, including the provision of complex care (e.g., symptom management, medication administration and controlling medical devices), organisational tasks (e.g., coordinating care and navigating insurance matters), making sound decisions while managing risks (e.g., weigh arguments and risk and consider alternatives) and managing family life with a child suffering from severe illness (e.g., embed their situation in a life as normal as possible and weigh the needs of other family members).[8] In addition, it has been reported that parents have to adapt their housing situation and manage substantial home modifications because of their child's medical complexity, including room reconfigurations and modifications (e.g., modifying the child's bedroom and the family's bathroom, widen doorways and installing ramps).[51,52]

The multiple roles and responsibilities, time-consuming and complex caregiving tasks, as well as the many life-altering changes that come with a child's LLC have an enormous impact on the life of families; for example, parents may face considerable financial and employment-related implications.[9,48-50] Overall, the burden of caregiving that parents of children with LLCs experience are manifold.[48] The following subchapter describes the multiple burdens that families of children with LLCs face, including their financial burden.

1.3.3 Caregiver burden

Navigating and managing the practical aspects of their child's LLC, while at the same time coping with the emotional impact, places parents at an increased risk of caregiver burden.[6,14,53] Caregiver burden—'*the level of multifaceted strain perceived by the caregiver from caring for*

a family member and/or loved one over time'[54,p.442]—is multidimensional and encompasses physical, psychological, emotional, social and financial aspects.[55]

Physical burden

In their review, Hartley et al.[50] drew attention to the impact of parenting a child with an LLC on parents' physical health. They noted that several studies reported parents' health to be directly and negatively impacted by taking care of their child and that these health effects evolve over time.[50] Physical pain, in particular back-pain, was one of the most common negative health impacts reported by parents.[50] They are likely a result of ongoing caregiving demands, including the frequent lifting of the child and/or medical equipment.[6,14] In addition, parents' health was found to be negatively affected by sleep deprivation due to a child's night-time seizures, abnormal sleep patterns and night-time care demands.[50] Mothers of children with LLCs also showed a higher incidence of cardiovascular disease, hypertension and type 2 diabetes compared to mothers of children with no long-term conditions.[13] This study furthermore found that mothers of children with LLCs are more hesitant to make their own health a priority, making them less likely to visit a general practitioner.[13]

Psychological and emotional burden

In studies exploring the psychological and emotional burdens associated with parenting a child with an LLC, the feeling of uncertainty has been highlighted repeatedly.[56] Parents reported to be negatively affected by the unremitting uncertainty emerging from a lack of control and the unpredictability of the progression of the child's illness, for example, regarding symptom severity, success or failure of treatment and considerations of life and death.[12,56] Throughout their child's course of illness, parents are likely to suffer profound distress,[57,58] anxiety,[7,13,56] and depression.[7,13] In addition, parenting a child with an LLC is a deeply emotional journey that can leave parents overwhelmed and emotionally exhausted.[5,53] Parents were found to experience an overwhelming emotional struggle around knowing that they do the best they can; their feelings of responsibility, frustration, losing control and failure; high levels of stress and the feeling of being socially isolated.[5] A study on the experiences of affected fathers found that emotional burden is especially high during times of intensive treatment, such as medical emergencies and invasive procedures and at the time of their child's death.[53]

Social burden

Socially, parents of children with LLCs experience difficulties in keeping connected and maintaining their social and spousal relationships, with some parents reporting estrangement and isolation from their social circles.[6,10,12,53,59] For instance, interviews with affected fathers showed that because of their demanding roles in caregiving, relationships with relatives and friends were negatively affected.[12] An Australian study about the experiences of parents of children with LLCs found that their child's often fragile and unpredictable condition kept them from participating in activities outside of the home.[6] Parents also reported shifts in their social circles, for example, connecting with other parents of children with LLCs via social support groups.[12] Additionally, having a child with an LLC was found to profoundly affect parents' spousal relationship.[10,53,59] While some parents reported that their relationship became practical, distant and less intimate because of the demanding and time-consuming caregiving tasks,[12,53,59] other parents' relationships became stronger.[10,53,59] They noted that a strong and supporting relationship was key to cope with the difficult experience.[10,53,59]

Financial burden

To measure the financial burden endured by families of children with LLCs, illness-related costs are commonly categorised as direct costs, that is, OOP medical and non-medical expenses, and indirect costs, that is, forgone employment-related income loss (see **Chapter 2** for more information).[60,61] While research on the illness-related costs incurred by parents remains limited for most LLCs, a more extensive body of literature is available in paediatric oncology.[16,17,62-79] These studies reveal that families of children with cancer face a wide range of adverse economic consequences.[16,17,62-79] For instance, studies from the United Kingdom,[65] Canada[16,66] and Australia[67,68] indicate that families incur considerable OOP non-medical expenses for travel and transport, accommodation, food, communication and childcare. While these costs were also considerable for families of children with cancer in India[69,70] and Colombia,[71] affected families in these countries also incurred high OOP medical expenses because of insufficient insurance coverage.[70] High OOP medical expenses for inpatient care, outpatient care and pharmaceuticals were also reported in a study on the cost of childhood cancer conducted in the United States.[64]

Regarding indirect costs, studies from the United States,[72-75] Canada,[76] Sweden[77] and Denmark[78] showed that parents, mainly mothers, of children with cancer experience substantial work disruptions and a loss of income. For example, a study conducted in the United States

found that 25% of participating families lost more than 40% of their annual household income due to illness-related work disruptions.[73] Moreover, in a Canadian study, 64% of mothers and 16% of fathers reported to have left their job after their child's cancer diagnosis.[76] And in Denmark, a study showed that even 10 years after diagnosis, mothers of children with cancer were more likely to not work or have a lower income in comparison to mothers with children without cancer.[78] Nevertheless, in a study conducted in Sweden, it has also been shown that social benefits largely compensate for the loss of income related to employment disruptions associated with childhood cancer.[79] On the other side, in countries with lower social benefits, such as India, income loss was found to substantially add to the costs incurred by affected families.[70] In this study, 72% of participating families reported illness-related income loss due to the employment impact of childhood cancer.[70]

A limited number of studies have examined the financial burden borne by parents of children with LLCs other than cancer, including cerebral palsy,[80-82] congenital heart disease[83,84] and muscular dystrophy.[85] In studies on families of children with cerebral palsy conducted in Nigeria[80] and Malaysia,[81] OOP medical expenses represented the highest costs, followed by OOP non-medical expenses and indirect costs. In Australia, a study showed that families' direct costs were highest for home modifications, travel and transport, splints and orthoses, childcare, equipment (e.g., for toileting and bathing) and medical technology.[82] In addition, a study in Denmark showed that forgone employment and lower income were more evident in mothers of children with cerebral palsy compared to mothers of children without cerebral palsy.[86] Regarding children with congenital heart disease, a study in the United States found that families of these children were more likely to spent >$1'000 (USD) a year on direct costs compared to those of children with special healthcare needs without congenital heart disease.[83] A study conducted in Nigeria found that families of children with congenital heart disease spent on average 26% of their income on direct costs, mainly for medicines and transportation.[84] Lastly, a study conducted in the United States showed that children with muscular dystrophy were more likely to have a family member who reported financial problems or reduced or stopped employment compared to families of children with special healthcare needs without muscular dystrophy.[85]

Nevertheless, irrespectively of the specific LLC, little is understood about the costs associated with illness-related events that parents have to navigate throughout their child's course of illness, for example, a child's hospitalisations or even its death. Evidence from cross-sectional studies in general paediatrics suggests that childhood hospitalisations are associated with

parents' missed working hours and increased OOP non-medical expenses arising from transportation and childcare.[87-89] Due to frequent and lengthy hospitalisations,[26,30,37-41] these effects may be even more pronounced in parents of children with LLCs. Likewise, the death of a child likely has considerable employment implications for parents. A qualitative study investigating the impact of bereavement, including the loss of a child, on the employment of grieving family members suggests that effects include increased absence from work, career disruptions, job resignation and employment termination.[90] In addition, a child's funeral expenses were found to be a major expense for families.[91]

Overall, it should be noted that the financial burden experienced by families of children with LLCs likely varies depending on national social security systems, including healthcare insurance. In Switzerland compulsory health insurance shields children and their families from incurring (high) OOP medical expenses, reducing their risk of experiencing financial burden.[92] In addition, a wide array of other social insurances and benefits offer protection against financial adversity (e.g., disability insurance, income compensation allowances, unemployment insurance and family allowances).[93]

1.4 Conceptual framework

1.4.1 Conceptual model of caregiver burden

To guide this research, a conceptual model of caregiver burden in parents of children with LLCs was developed. The conceptual model seeks to establish the underlying structures of the relationship between a child's LLC and the physical, psychological, emotional, social and financial burdens experienced by parents. The model was conceptualised by drawing from Reuben Hill's[94] prior contribution, extensions thereof,[95-101] and the research conducted by Raina et al.[102]

In 1958, Hill proposed a model—*the ABC-X Model of Family Stress*—to analyse factors explaining the relationship between a life-altering event, i.e., a stressor event, and its manifestation, i.e., crisis within a family.[94,103,104] The model consists of four parts: a stressor event (A), mediating factors (B and C) and an outcome (X).[94,103,104] A stressor event can be any condition or event that has the potential to deeply impact a family.[94,103,104] The mediating factors refer to a family's available resources and its perception of events, which both may mediate the manifestation of the outcome of the stressor event.[94,103,104] Thereby, the outcome is a consequence

of the demands/costs that a stressor event places on a family, for example, emotional demands, financial costs or adverse health effects in family members.[94,103,104]

Over the years, several extensions of Hill's[94] *ABC-X Model* have been developed, which all added new aspects to the original model. The *Double ABC-X Model of Family Stress and Adaptation* added time as a factor, suggesting that the manifestation of the stressor event is an evolving process.[95,96,103,104] By taking time into consideration, the model incorporates the effects of stressor-related events that follow the initial stressor event.[95,96,103,104] The manifestation process is then characterised by a pile-up of demands, that is, a cumulation of effects or costs over time.[95,96,103,104] The concepts of adjustment and adaption were added in both the *Double ABC-X Model of Family Stress and Adaptation* and the *Family Adjustment and Adaptation Response Model*.[95-97,103,104] The models suggest that families adjust (minor changes) or adapt (considerable, disruptive changes) in response to the manifestation of a stressor event.[95-97,103,104] While the *Model of Family Response to a Child's Chronic Illness or Disability* drew attention to the risk factors potentially aggravating the manifestation of the stressor event,[98] the *Resiliency Model of Family Stress, Adjustment and Adaptation* proposed family resiliency as a protective factor alleviating the demands placed on families.[99,103,105] The latter model suggests that certain family characteristics and properties increase a family's level of resistance to disruptions.[99,103,105] A study applying the model to childhood cancer identified several factors increasing a family's resilience, relating to the internal strength of the family, external support networks and the family's appraisal of events.[106] The *Contextual Model of Family Stress* is a further extension of Hill's[94] original *ABC-X Model*.[100,101,103] It proposes that the process of the manifestation of the stressor event occurs within the context of value, belief, cultural, economic and political systems.[100,101,103]

In designing the conceptual model, consideration was also given to Raina et al.'s[102] multidimensional *Model of Caregiving Process and Caregiver Burden Among Paediatric Population*. By presenting several caregiving-related factors, such as child characteristics, caregiver strain and self-perception, the model illustrates how parental caregiving can cause adverse psychological and physical health outcomes in families of children with disability.[102] Additionally, they highlight that caregiving occurs within a family's social and economic context, including the setting in which caregiving takes place.[102] The developed conceptual model is presented in Figure 1.

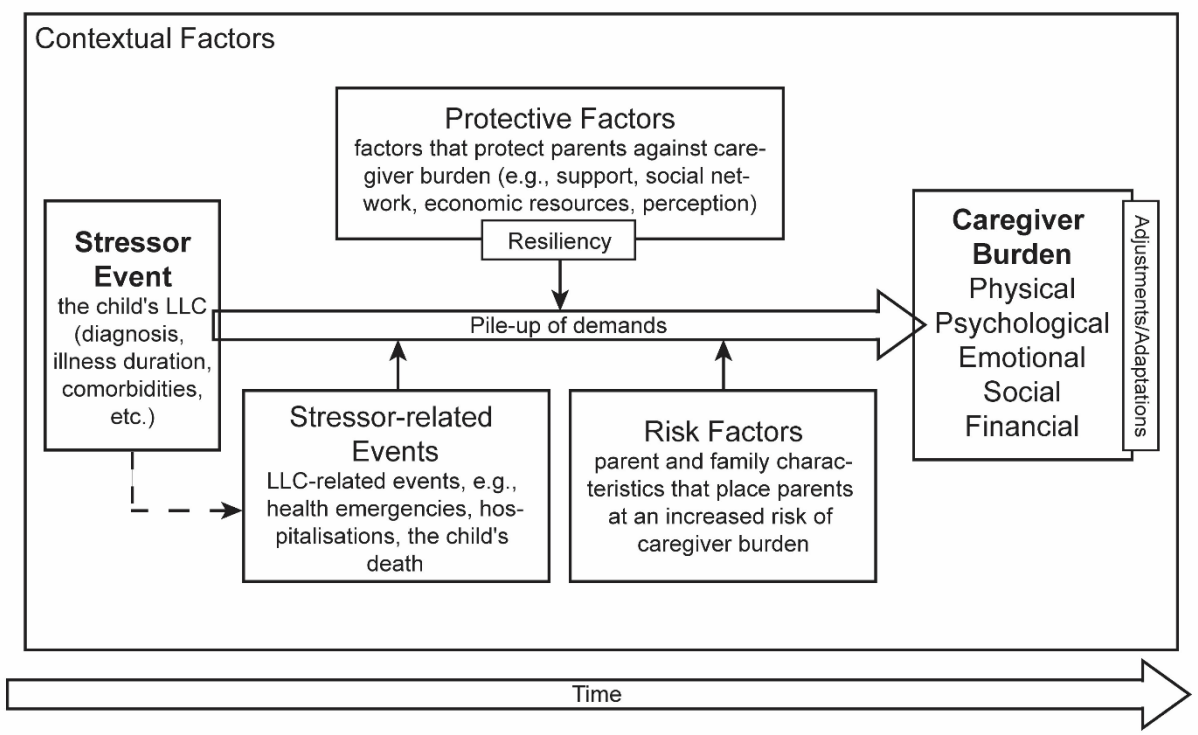

Figure 1: Model of Caregiver Burden in Parents of Children with LLCs.

The developed conceptual model presents several key components, each playing a crucial role in the manifestation of caregiver burden in parents of children with LLCs. The central idea is that the LLC of a child, i.e., the stressor event, and LLC-related events, i.e., stressor-related events, place demands on parents that pile-up over time and manifest in physical, psychological, emotional, social and financial caregiver burdens. In other words, parents may face caregiver burden due to increasing illness-related demands, which may be even further exacerbated by LLC-related events, such as hospitalisations or the death of a child. For example, parents may incur extra travel costs when visiting their hospitalised child daily.

Risk and protective factors influence the likelihood of parents experiencing caregiver burden. While certain parent and family characteristics, such as single parenthood, may place parents at an increased risk of caregiver burden, protective factors may shield parents. This may include resiliency-increasing factors such as support from care programmes (e.g., SPPC), support from relatives and friends, a family's economic resources or parents' perception of events.

Adjustments and adaptations constitute a way for parents to cope with caregiver burden. For example, parents could adjust their caregiver roles by reassessing and redistributing

responsibilities or adapting their living situation by moving closer to relatives to strengthen their social support network.

Overall, the process of caregiver burden manifestation in parents of children with LLCs occurs within the wider socio-cultural and socio-economic context. Regarding financial burden, social security and healthcare system characteristics may be of particular relevance.

1.4.2 The concept of financial burden

Financial caregiver burden can be understood as an adverse economic spillover effect or externality that arises from an illness or health condition, for example, the LLC of a child, and extends beyond the directly affected individual to family caregivers, for example, the child's parents.[107-109] Financial burden has both an objective and a subjective dimension.[110,111] While objective financial burden refers to the monetary costs incurred by patients or caregivers, subjective financial burden describes the subjective appraisal of those costs.[110] Subjective financial burden is commonly measured as financial distress or financial hardship.[110,112] Objective and subjective financial burdens have also been described as material and psychological financial burden, respectively.[111] While recognising the lack of evidence on both the objective and subjective financial burdens experienced by families of children with LLCs, this dissertation's focus is on objective financial burden. An enhanced understanding of subjective financial burden, requires first a better understanding of the preceding monetary costs, that is, the objective financial burden.

The cost-of-illness methodology provides a framework for measuring the objective financial burden arising from disease.[60,61] It facilitates the assessment of monetary costs incurred by patients or caregivers through the definition of assessment approaches and cost categories, including direct costs, that is, OOP medical and non-medical expenses, and indirect costs.[60,61] The concept of cost-of-illness will be further detailed in the introduction of **Chapter 2**.

While the cost-of-illness methodology provides a generic framework for quantifying financial burden,[60,61] distinct sets of established cost measures for assessing the costs incurred by families of children with LLCs are largely lacking. Prior attempts to synthesise existing evidence of the costs incurred by families of children with LLCs encountered major difficulties due to discrepancies in definitions, terminology and a general lack of standardisation of relevant cost measures.[17,62,63] Problematically, the absence of consensus in quantifying the costs incurred by

affected families not only hinder accurate comparisons of research findings,[17,62,63] but may also raise the risk of overlooking certain costs, likely compromising the comprehensiveness of financial burden assessments.

1.5 Specialised paediatric palliative care

1.5.1 Model of care

Paediatric palliative care (PPC) is advocated as a standard of care for children with LLCs and their families.[18,113-115] It is the active holistic care of children with serious health-related suffering because of severe illness, i.e., LLCs.[116,117] PPC includes the '*prevention, early identification, comprehensive assessment, and management of physical issues, including pain and other distressing symptoms, psychological distress, spiritual distress, and social needs*'[116,p.761] and aims to improve and maintain the best possible quality of life for children and their families.[116,117] Optimally, it is introduced at the time of diagnosis and continues throughout a child's life and into a family's bereavement.[18]

Ideally, PPC is provided by specialised, multi-professional teams, that is, SPPC teams, working exclusively (dedicated) in SPPC.[18] While basic PPC services may be provided by medical specialists in a fragmented manner, SPPC teams provide an additional, more comprehensive and specialised layer of care that complements the care provided by medical disciplines and in different settings (Figure 2).

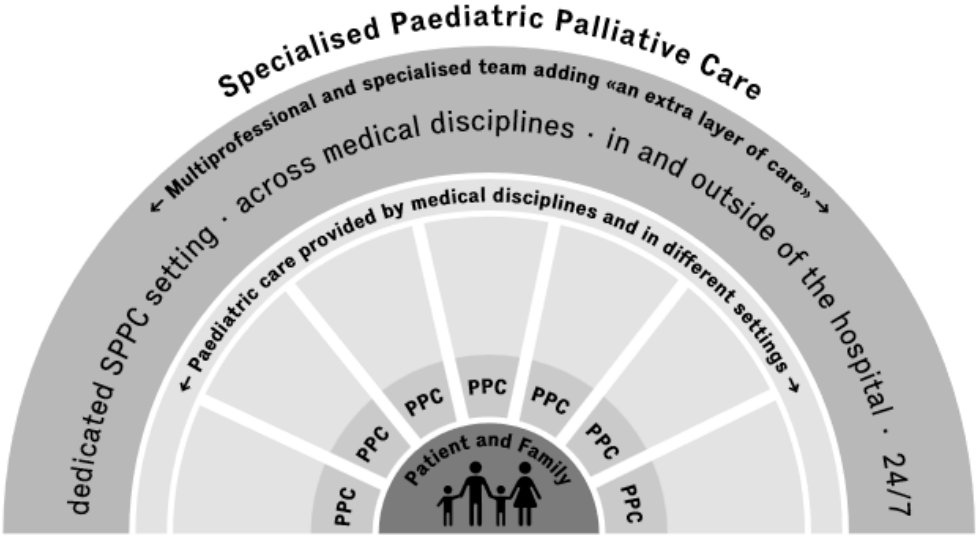

Figure 2: Model of Specialised Paediatric Palliative Care. (Illustration by SPhAERA team.)

Multi-professional SPPC teams consist of physicians, nurses, psychologists, therapists, social workers and other professionals specialised in PPC.[18] They work fully engaged and dedicated in SPPC and provide specialist care and support through consultancies inside and outside of hospitals, for example, in a family's home or in other healthcare facilities, and are available continuously 24 hours a day, 7 days a week, 365 days a year.[18] SPPC is commonly delivered using a consultative model of care, as it enables the provision of SPPC alongside the primary care provided by medical specialists, therapists and other professionals.[18] Additionally, the consultative model of care ensures the availability of SPPC in all diverse settings (inside and outside of the hospital), where a child and its family receive care and support.[18]

1.5.2 Supporting parents

Providing support to family members of children with LLCs is an integral part of SPPC.[18] In addition to the child's siblings, grandparents and other close family members, this specifically pertains to parents.[18] By addressing parents' multiple support needs, SPPC teams help them navigate the demanding caregiving responsibilities and ease the caregiver burden that comes along with it.[18,118-120] It has been suggested that SPPC teams can support parents of children with LLCs by providing psychological and emotional support (e.g., psychological counselling),[121,122] facilitating decision-making processes,[121] coordinating care with other specialities and professionals,[121,122] initiating spiritual and religious support appropriate to a family's beliefs and practises,[18] supporting parents in maintaining their own health (e.g., increasing awareness and organising respite care),[121,122] supporting parents in maintaining their social roles (e.g., providing information about support groups),[122] and by providing relevant information on available hospital and community resources.[122] Thereby, the support provided to parents should be characterised by open communication, active involvement and a trusting relationship.[18,121,122]

Furthermore, it has been suggested that SPPC teams can support parents by addressing their financial support needs.[18,122] Strategies to mitigate the financial burden endured by parents may include the provision of relevant information, referral to a financial advisor and the involvement of a social worker.[122-124] Ideally, a social worker specialised in PPC is part of the multi-professional SPPC team.[123] As outlined in a recent review, providing support regarding financial, legal and work-related matters is one of the core competencies of social workers specialised in PPC.[123] This encompasses tasks such as an assessment of a family's available resources;

informing parents about local, state and federal financial support programmes; help with application processes and refer parents to external advisory services, such as insurance specialists, legal experts or financial advisors.[123]

Although there is a lack of literature on best practice strategies to address families' financial support needs in PPC, a study in general oncology identified three models of financial support provision, which may also apply to PPC: financial counselling, financial advocacy and financial navigation.[125] In the financial counselling model, healthcare professionals refer patients to a hospital's internal or external financial counsellor.[125] In contrast, the financial advocacy model refers to a more direct support by healthcare professionals, which supports patients and their families by providing information, helping with application processes and giving advice regarding legal and work-related matters.[125] While both of these modes are reactive, the financial navigation model is proactive.[125] In this model a dedicated team member directly addresses the financial support needs of patients and their families, while also staying up-to-date with the patient's medical treatment and care.[125] This enables a proactive provision of support, for example, proactively optimising a patient's insurance coverage.[125]

1.5.3 The Swiss context

Although basic PPC services are provided by medical specialists throughout Switzerland, the availability of SPPC programmes varies across different regions.[126,127] Where SPPC is available, it is provided by hospital-based consultative SPPC programmes.[128] These programmes are located at (university) children's hospitals and provide support and care via consultancies as and where necessary—both inside and outside of hospitals—to patients, their families, primary care teams and other healthcare professionals.[128] However, even existing programmes vary considerably with regards to their personnel and financial resources.[126,127] As a consequence of the lack of nationwide coverage, not all children with LLCs and their families currently have access to SPPC.[126,129] Moreover, for children and families enrolled in an SPPC programme, provision of care and support is likely constrained by resource limitations.[130] For instance, some programmes currently lack the resources to deliver support and care beyond the hospital setting, for example, in long-term care facilities or a family's home.[130] These shortcomings in SPPC access and provision may, in part, be due to inadequate service reimbursement and funding constraints.[126,127,131] A lack of awareness in politics, policy-making and insufficient educational and training opportunities may constitute further barriers.[131] Nevertheless,

funding-related constraints in SPPC access and provision may have considerable consequences for families, as they may not receive the care and support they need in order to cope with the many burdens that come with a child's LLC.

Although **Chapter 4** provides detailed insights into the current funding of hospital-based consultative SPPC programmes in Switzerland, it should be recognised that in the Swiss healthcare system, resources to pay for healthcare services, including SPPC, are collected mostly through compulsory insurance premiums and taxes.[92] In addition, patients utilising insured healthcare services are required to contribute through cost-sharing mechanisms, including deductibles and co-payments.[92] And while patients under the age of 18 are exempt from deductibles, their families are still liable for co-payments.[132] However, these co-payments are capped at a relative low level.[132]

1.6 The SPhAERA study and beyond

This dissertation's investigation of financial burden in families of children with LLCs (**Chapters 2** and **3**) is conducted within the frame of the 'Specialised Paediatric PAlliativE CaRe: Assessing family, healthcare professionals and health system outcomes in a multi-site context of various care settings (SPhAERA)' study. The SPhAERA study is a comparative effectiveness study evaluating a Swiss SPPC programme with respect to its potential to improve patient-, family-, health professional-, and healthcare-related outcomes. The study was conducted between November 2019 and May 2023 at four Swiss (university) children's hospitals, i.e., Aarau, Basel, Bern and Zurich. While this dissertation focuses on the analysis of family-level financial and employment data, other data collected in the SPhAERA study, such as quality of life, are analysed in separate sub-studies. Detailed information on the SPhAERA study can be found in the SPhAERA study's protocol in the **Appendix**.

In addition, recognising that funding and reimbursement issues are likely a major barrier to the implementation and provision of SPPC in Switzerland, data on the current funding of hospital-based consultative SPPC programmes were collected as part of this dissertation. This data collection took place independently from the SPhAERA study. **Chapter 4** provides further details on the related data collection and analyses procedures.

1.7 Research gap and rationale

In recent decades, the prevalence of LLCs in children has risen steadily.[1-3] This increase has profound implications, not only for affected children but also for their parents, who may experience substantial caregiver burden.[5-7] While a growing body of literature has explored the physical, psychological, emotional and social burdens on parents, evidence on the financial burden remains limited. However, robust evidence is critical to develop and utilise effective, evidence-based strategies aimed at the prevention and relief of financial burden in families of children with LLCs, such as delivered by SPPC programmes.

Today, there are several factors that hinder the development and implementation of effective financial support programmes. First, inconsistencies in cost measures, definitions and methodological approaches used across studies that quantify financial burden in families of children with LLCs hinder the interpretation and comparability of research findings.[17] These inconsistencies are likely constraining efforts to derive meaningful conclusions regarding the magnitude of financial burden faced by affected families. As a result, the development of effective support programmes is likely hampered.

Second, the existing evidence base is predominately cross-sectional in nature, lacking longitudinal studies quantifying the monetary costs borne by parents of children with LLCs over time. Particularly, this applies to the financial and employment implications of LLC-related events, such as hospitalisations or the death of a child. To my knowledge, no LLC-specific longitudinal studies investigating these effects have yet been published. However, such evidence is needed to better understand the manifestation of financial burden over time.

Third, beyond these challenges, a critical issue that needs attention is the funding landscape for SPPC programmes. In Switzerland, funding issues may constitute a major barrier to the provision of SPPC. Inadequate funding may restrict SPPC's accessibility and provisions, impeding its widespread availability. Consequently, there is a risk that families of children with LLCs may not receive the comprehensive support they require, including support aimed at addressing their financial support needs.

Nevertheless, parenting a child with an LLC is an enormously challenging and demanding journey, that may come with profound financial burden due to high illness-related costs. By bridging the outlined gaps in evidence, this dissertation hopes to pave the way for more effective and targeted strategies to address the financial support needed by parents of children with

LLCs. Offering considerate support that protects families from adverse illness-related financial effects will improve the quality of life for both parents and their children.

1.8 Study aims

This dissertation includes the following aims:

(I) To provide a comprehensive overview of existing cost indicators and outcome measures used to quantify financial burden in families of children with LLCs and to provide clear recommendations for future assessments of financial burden (**Chapter 2**).

(II) To explore how a child's hospitalisation influences the family's OOP non-medical expenses as well as the parents' income and employment over a 330-day period and to assess changes in parental income and employment over the first 300 days of bereavement (**Chapter 3**).

(III) To investigate the funding of SPPC in Switzerland by developing a conceptual model visualising the current funding of SPPC programmes and by identifying obstacles to and priorities for funding these programmes sustainably (**Chapter 4**).

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Chapter 2: Measuring Financial Burden in Families of Children Living With Life-Limiting Conditions: A Scoping Review of Cost Indicators and Outcome Measures

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2.1 Highlights

- Families of children suffering from life-limiting conditions face enormous challenges, including substantial illness-related financial burden. Nevertheless, inconsistencies in outcome measures, definitions and methodological approaches limit the interpretation and comparability of research findings, hindering the development and implementation of effective support policies.
- This study provides a comprehensive overview of exiting cost indicators and outcome measures used to measure financial burden in families of children with life-limiting conditions. On the basis of our findings, we developed a framework for measuring out-of-pocket expenditures and provided clear recommendations for assessing income loss, opportunity costs and financial support. We hope that our findings will help to consistently and precisely quantify the true weight of families' financial burden.
- Evidence on the true prevalence and severity of families' financial burden is needed to develop and implement effective healthcare policies. Our review suggests that not all key components of families' financial burden were consistently measured across studies. We hope that this article's framework for measuring out-of-pocket expenditures and our recommendations will contribute to more consistent and comprehensive measurements of financial burden, providing critical information for evidence-based policy making.

2.2 Abstract

Objectives

This study aimed to provide a comprehensive overview of cost indicators and outcome measures used to measure financial burden in families of children with life-limiting conditions.

Methods

A scoping review methodology was used to map the existing literature and provide an overview of available cost indicators and outcome measures. Key medical, economic and scientific databases were systematically searched to identify relevant articles published in 2000 or later.

Results

The database search yielded 7194 records, including 30 articles eligible for final inclusion. Retrieved cost indicators and outcome measures fell into three broad categories: direct costs,

indirect costs and financial support. No study comprehensively assessed all three categories. Cost indicators used to measure direct costs were grouped into 5 medical and 11 non-medical out-of-pocket expenses categories, of which 5 were commonly assessed (i.e., treatment and diagnostics; travel and transport; accommodation; food; childcare and home help). Half of the reviewed studies included assessments of indirect costs, most commonly estimating work-related income loss by evaluating employment disruptions. Assessments of opportunity costs arising from informal caregiving and of financial support were rarely included.

Conclusions

Current estimates of the financial burden faced by families of children with life-limiting conditions are inconsistent and often incomplete, likely resulting in severe underestimations of the costs these families incur. We hope that the framework presented in this paper will contribute to a more comprehensive assessment of illness-related financial burden and help guide future policies in this area.

2.3 Introduction

Suffering from life-limiting conditions (LLCs) is a traumatic experience not only for the directly affected children but for their entire families.[1] Globally, the number of children living with LLCs has increased rapidly over the last two decades. In Queensland, Australia, estimates indicate an increase in LLC prevalence from 35.2 to 43.2 per 10'000 population between 2011 and 2016.[2] In England, UK, prevalence increased from 26.7 to 66.4 (per 10'000) between 2001/2002 and 2017/2018.[3] Although advances in life-extending medical care and technology can partially explain these figures,[4,5] changes in medical coding practice and recording may also have had an effect.[3]

According to Together for Short Lives[6], LLCs encompass four groups of conditions: (1) conditions for which curative treatment may be feasible but can fail, for example, cancer or irreversible organ failure; (2) conditions in which premature death is inevitable but in which there may be long periods of intensive treatment, for example, Duchenne muscular dystrophy; (3) progressive conditions without curative treatment options, for example, severe metabolic conditions; and (4) irreversible but non-progressive conditions causing severe disability creating susceptibility to health complications and likelihood of premature death, for example, cerebral palsy.

Caring for a child with an LLC is a profoundly difficult and dramatic experience for affected families. Persistent stress, anxiety, feelings of fear and deteriorations of family life are commonly reported.[1,7,8] Symptoms of distress and anxiety can be further compounded by financial stressors, including not only non-reimbursed expenses for hospitalisations, medications and equipment,[7] but often conflicts between employment obligations and childcare responsibilities.[8]

Considerable research has examined families' financial burden in paediatric oncology. Systematic reviews of the cost of childhood cancer from a family perspective have identified a large variety of adverse economic consequences,[9-11] including substantial out-of-pocket (OOP) expenses[10] as well as parental employment disruptions and income loss.[9] Nevertheless, because few studies have reported illness-related costs in a comprehensive and comparable manner, it is difficult to exploit published data for research and policy making.[9-11]

Measuring financial burden

Cost-of-illness studies provide a framework for measuring the financial burden of disease to families.[12,13] These studies traditionally distinguish between direct and indirect costs and, if applicable, also adjust for financial support received.[12,13] Direct costs, that is, OOP expenses directly incurred by affected families, are commonly divided into medical and non-medical expenses.[12] OOP medical expenses, that is, those directly related to the consumption of healthcare resources, include co-payments, deductibles, co-insurances and all direct charges not covered by formal payers.[12] OOP expenses not related to the consumption of healthcare resources, but nevertheless necessitated directly by health conditions, such as travel costs to and from hospitals, are classified as non-medical.[12] Indirect costs include changes in productivity and work, for example, when parental caregiving responsibilities require changes in employment status (leaving or reducing work) that cut income.[12,13] For informal caregivers, work-related income loss and opportunity costs due to forgone earnings are common indirect costs.[13] Whether costs are direct or indirect, medical or non-medical, financial support from both governmental and non-governmental entities may be available to mitigate the resulting financial burden.

Although cost-of-illness study designs provide a generic framework for measuring financial burden, distinct sets of established indicators and cost measures for measuring direct and indirect costs are largely lacking. In addition, previous reviews found substantial inconsistencies in these studies' definitions and terminology and a general lack of standardisation of relevant

outcome measures.[9-11] For example, indicators and outcome measures differed depending on whether studies used micro-costing or general estimates.[11] Problematically, the general lack of consensus on cost indicators and outcome measure classifications causes inconsistencies in reporting limiting the extent to which findings can be compared across studies.[9,10] Such reporting differences appear particularly large regarding financial support received by families.[9]

The purpose of this scoping review was to provide a comprehensive overview of existing cost indicators and outcome measures used to measure financial burden in families of children with LLCs. To increase standardisation and consistency, we also aimed to categorise retrieved cost indicators and outcome measures, along with providing detailed information on how they were assessed. We expect that reducing inconsistencies in this way will facilitate the development and implementation of effective healthcare policies supporting affected families by reducing adverse financial consequences.

2.4 Methods

2.4.1 Study design

This scoping review was conducted following the five-step methodological framework (i.e., research question identification; study identification; study selection; charting of data; collating, summarising and reporting results) proposed by Arksey and O'Malley[14] and developed by Levac et al.[15] By including studies of diverse designs and methodologies, scoping reviews can provide a broad descriptive overview of the nature and characteristics of available research and its findings.[14,15] Therefore, they provide an ideal framework for mapping, consolidating and disseminating evidence concerning cost indicators and outcome measures used to measure a topic as complex as financial burden.

2.4.2 Search strategy

For advice on designing our database search strategies, we consulted an experienced librarian. The resulting search strategies, including search terms and subject headings, are available as Supplemental Materials. The Boolean operators AND and OR were used to combine search terms and subject headings. We searched titles and abstracts in the MEDLINE and CINAHL medical databases, the EconLit economic database and the Scopus science database. Subject headings were added when searching MEDLINE, CINAHL and EconLit using the EBSCOhost

research platform. Grey literature was located using Google Scholar web searches. The database search was performed first between May 29 and June 3, 2019, then again on March 18, 2020 to find any newly published articles.

2.4.3 Eligibility criteria

Published articles were included if they reported on financial burden among families of children with LLCs, including any identifiable direct and indirect illness-related costs. To account for recent advances in research methodology and changes in health policy, we included all relevant studies published in 2000 or later. No language restrictions were applied. Studies where parents of children with LLCs were not the main focus (e.g., those including non-life-limiting chronic illnesses such as asthma, allergies or migraines) were excluded. Studies were also excluded if they assessed perceived rather than economically quantified burden.

2.4.4 Study selection

Search results were stored and managed via EndNoteX9™ reference software (Clarivate Analytics, London, UK). After removing duplicates, two study team members (T.M. or A.K.G. and S.M.) independently screened titles and abstracts, then independently assessed the full text of those retained. At each selection stage, any disagreements were resolved via discussions involving a third team member (K.Z.).

2.4.5 Data charting and reporting

We extracted data regarding study characteristics, including study design, participants, assessed outcomes, recall periods and data collection mode, as well as authors' names, year of publication and country of origin (Table 1). Cost indicators and outcome measures were grouped into three categories; direct costs, indirect costs and financial support. Where studies reported direct costs, data were further grouped into OOP medical and non-medical expenses categories (Table 2). Data on indirect costs (income loss, opportunity costs) and financial support are provided respectively in Tables 3 and 4. All indicators and their categorisations were discussed within the study team.

Because this study's purpose was to provide structured evidence regarding previously reported cost indicators and outcome measures, included studies' quality was not assessed. As a

reporting guideline, we followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) extension for scoping reviews (PRISMA-ScR).[16]

2.5 Results

2.5.1 Study selection

Our database search yielded 7194 records. After removal of duplicates, screening of titles and abstracts, full-text assessments and a grey literature search, 30 articles reporting on 28 studies qualified for inclusion. Figure 1 illustrates the selection process using a PRISMA flow chart.

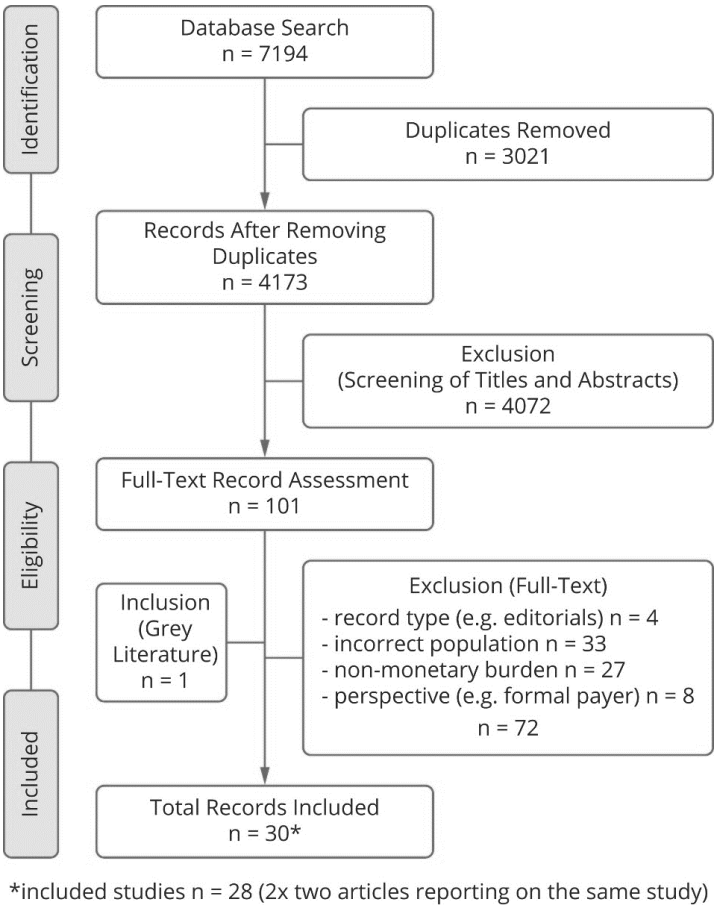

Figure 1: PRISMA flow diagram of study selection. PRISMA indicates Preferred Reporting Items for Systematic Reviews and Meta-analyses.

2.5.2 Characteristics of included studies

Of the 28 studies covered by the 30 included articles, nine were conducted in the United States,[17-25] with a tenth, cross-national, study involving both Australian and US participants.[26]

Additionally, three each were conducted in Australia[27-29] and India,[30-32] two each in Denmark[33,34] and Nigeria,[35,36] and one each in Canada,[37,38] Colombia,[39] Finland,[40] Italy,[41] Malaysia,[42] New Zealand,[43] Sweden,[44,45] and the UK.[46]

All included studies were observational, but used various data collection modes and recall periods. Sixteen used a cross-sectional study design,[18-22,25-30,36,39,42,43,46] collecting data with paper-pencil questionnaires or phone-administered surveys,[18-22,25,27-29,36,39,42,43,46] or combining face-to-face interviews with a self-report questionnaire.[26,30] Of these 16, seven studies assessed expenses retrospectively, with a recall period of 12 months.[18-21,25,27,42]

Twelve studies used a longitudinal study design,[17,23,24,31-35,37,38,40,41,44,45] collecting data either via subject reports, that is, face-to-face interviews,[24] repeated questionnaire surveys[35,40] or cost diaries,[17,31,32,37,38] or by linking data from administrative registries and medical claims databases.[23,33,34,41,44,45] Interview and questionnaire surveys were conducted for 3,[35] 6,[24] or 15 months,[40] including either two[24,40] or three[35] assessment timepoints. Families were asked to record expenses in cost diaries for a maximum of 12 weeks.[32] Three population-based studies, two conducted in Denmark and one in Sweden, linked data from various government-administered registries,[33,34,44,45] which allowed assessment periods ranging from 8[44,45] to 28 years.[33]

Seventeen studies examined the financial impact of paediatric cancer[22,24-28,30-32,34,37-41,43-46] and six the costs related to cerebral palsy.[17,19,29,33,36,42] Three used samples of children suffering from congenital heart disease.[21,23,35] Ouyang et al.[20] surveyed families with children suffering from muscular dystrophy and Thomson et al.[18] examined a sample of children with complex medical conditions, including neurological and congenital conditions and paediatric cancer. Overall, included studies collected cost data during different phases of illness and treatment.

Cost indicators and outcome measures assessed in the included studies were grouped into three categories—direct costs (OOP medical and non-medical), indirect costs (income loss and opportunity costs), and financial support. No study comprehensively assessed all three categories. Eight studies assessed both direct and indirect costs,[17,27,28,30,37,38,40,42,43] with the remainder assessing only direct[18-23,29,31,32,35,36,39,46] or indirect[24-26,33,34,41,44,45] costs. Of the 15 reporting indirect costs, 14 assessed parental income loss[17,24-28,30,33,34,37,40,42-45] and two the opportunity costs of informal caregiving.[37,41] The extent to which OOP medical[18-21,23,27-29,31,32,35-38,40,42,43] and non-medical[17,22,27-32,35-40,42,43,46] expenses were measured varied across studies. Financial support was assessed in four studies.[27,31,43,46] All included studies' characteristics are presented in Table 1.

Table 1: Characteristics of included studies.

Autor(s), *country*	Design	Participants	Time horizon and data collection mode	Assessed outcomes				
				Direct costs		Indirect costs		Financial support
				Out-of-pocket medical	Out-of-pocket non-medical	Income loss	Opportunity costs	
Ahuja et al[32] (2019) *India*	Longitudinal	Families (n=11) of children (3-18 years) diagnosed with cancer	14-week: 2 weeks prior to diagnosis (questionnaire survey); 12 weeks following the diagnosis (cost diary)	X	X			
Badaru et al[36] (2019) *Nigeria*	Cross-sectional	Families (n=106) of children (1-11 years) diagnosed with cerebral palsy	Per hospital visit or 6 months retrospective, depending on the assessed cost item (questionnaire survey)	X	X			
Bona et al[25] (2014) *USA*	Cross-sectional	Families (n=71) of children (2-18 years) diagnosed with cancer	12 months (questionnaire survey)			X		
Bona et al[24] (2016) *USA*	Longitudinal	Families (n=99 in T1/ n=93 in T2) of children (median = 8.9 years) diagnosed with cancer	First interview within 30 days following diagnosis, second interview 6 months after diagnosis (repeated face-to-face interviews)			X		
Bourke-Taylor et al[29] (2013) *Australia*	Cross-sectional	Families (n=29) of children (2-12 years) diagnosed with cerebral palsy	Maximum period of 6 years following diagnosis (questionnaire survey)	X	X			
Cohn et al[28] (2003) *Australia*	Cross-sectional	Parents (n=100) of paediatric cancer patients (0.8-18 years)	21-5475 days following diagnosis (questionnaire survey)	X	X	X		
Dockerty et al[43] (2003) *New Zealand*	Cross-sectional	Parents (n= 237) of children (0-14years) diagnosed with cancer	30 days (questionnaire survey)	X		X		X

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Autor(s), *country*	Design	Participants	Time horizon and data collection mode	Assessed outcomes				
				Direct costs		Indirect costs		Financial support
				Out-of-pocket medical	Out-of-pocket non-medical	Income loss	Opportunity costs	
Dussel et al[26] (2011) *USA & Australia*	Cross-sectional	American (n=141) and Australian (n=89) parents of deceased childhood cancer patients	Annual income loss assessed at a median of 3.3 years (USA; telephone survey) and 4.4 years (Australia; face-to-face interview and paper-pencil self-report questionnaire) after death			X		
Eiser and Upton[46] (2006) *UK*	Cross-sectional	Parents (n=145) of children (1-20 years) diagnosed with any cancer before 16 years of age	Past-week spending over and above pre-illness expenditure (questionnaire survey)		X			X
Elhoff et al[23] (2018) *USA*	Longitudinal; register-based cohort study	Families (n=481) of children born with severe congenital heart disease	During the first 12 months of life or until death, if sooner (administrative claims register data)	X				
Fluchel et al[22] (2014) *USA*	Cross-sectional	Primary caregivers (n=354) of paediatric cancer patients (0-18 years)	Per clinic visit (questionnaire survey)		X			
Ghatak et al[31] (2016) *India*	Longitudinal	Families (n=50) of paediatric patients (1-12 years) with acute lymphoblastic leukaemia	During the first month of therapy, recording expenses on a daily basis (cost diary)	X	X			X
Heath et al[27] (2006) *Australia*	Cross-sectional	Families (n=56) of children (mean=7.6 years) diagnosed with cancer	12 months following diagnosis (questionnaire survey)	X	X	X		X
Hiyoshi et al[45] (2018) & Lindahl-Norberg et al[44] (2017) *Sweden*	Longitudinal; register-based cohort study	Families (n=20,091) of children (0-18 years) with (n=1899) and without (n=18192) cancer	Families were followed for up to 8 years starting from 1 year before the child's diagnosis (register data)			X		

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Autor(s), *country*	Design	Participants	Time horizon and data collection mode	Assessed outcomes				
				Direct costs		Indirect costs		Financial support
				Out-of-pocket medical	Out-of-pocket non-medical	Income loss	Opportunity costs	
Kamaral-zaman et al[42] (2018) *Malaysia*	Cross-sectional	Parents (n=74) of children (0-18 years) diagnosed with cerebral palsy	12 months (questionnaire survey)	X	X	X		
Lhteenmäki et al[40] (2004) *Finland*	Longitudinal	Families (n=21) of children (0.1-15 years) diagnosed with cancer	First survey 3 months after diagnosis, second survey 12 months later (repeated questionnaire survey)	X	X	X		
Mader et al[34] (2020) *Denmark*	Longitudinal; register-based cohort study	Parents of children with (n=12418) and without (n=125014) cancer	Follow-up period of 10 years starting 1 year before cancer diagnosis (register data)			X		
McClung et al[21] (2018) *USA*	Cross-sectional	Families (n=1956) of children (0-17 years) with congenital heart disease	12 months (telephone survey)	X				
Michelsen et al[33] (2015) *Denmark*	Longitudinal; register-based cohort study	Parents (n=21654) of children with (n=3671) and without (n=17983) cerebral palsy	Follow-up period of 28 years, starting 2 years before birth (register data)			X		
Ouyang et al[20] (2012) *USA*	Cross-sectional	Parents of children (0-17 years) with (n=112) and without muscular dystrophy	12 months (telephone survey)	X				
Pagano et al[41] (2014) *Italy*	Longitudinal; register-based cohort study	Families (n=917) of children and adolescents (0-19 years) diagnosed with cancer	3-year follow-up after diagnosis (register data and administrative health data)				X	

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Autor(s), *country*	Design	Participants	Time horizon and data collection mode	Assessed outcomes				
				Direct costs		Indirect costs		Financial support
				Out-of-pocket medical	Out-of-pocket non-medical	Income loss	Opportunity costs	
Rativa and Carreno[39] (2018) *Colombia*	Cross-sectional	Families (n=50) of children (1-14 years) with cancer	Monthly expenses (questionnaire survey)		X			
Sadoh et al[35] (2019) *Nigeria*	Longitudinal	Families (n=32) of children (0.5-5 years) with congenital heart disease	Monthly expenses for 3 consecutive months (repeated questionnaire survey)	X	X			
Schaible et al[19] (2018) *USA*	Cross-sectional	Families (n=744) of children (0-17 years) with cerebral palsy	12 months (telephone survey)	X				
Sneha et al[30] (2017) *India*	Cross-sectional	Families (n=70) of children (mean=7.8 years) diagnosed with acute leukaemia	During hospitalisation (questionnaire survey and face-to-face interviews)		X	X		
Thomson et al[18] (2016) *USA*	Cross-sectional	Families (n=167) of children (0-18 years) with complex medical conditions	12 months (questionnaire survey)	X				
Tsimicalis et al[37,38] (2012) & (2013) *Canada*	Longitudinal	Families (n=99) of children (0-18 years) diagnosed with cancer	1 week per month for 3 consecutive months (cost diary and face-to-face interviews)	X	X	X	X	
Vessey et al[17] (2017) *USA*	Longitudinal	Parents (n=52) of children (mean=11.5 years) diagnosed with cerebral palsy	During hospitalisation (average length of stay=9.36 days) (cost diary)		X	X		

2.5.3 Direct costs: out-of-pocket medical and non-medical expenses

Both OOP medical and non-medical expenses were reported for varying periods of time and to different extents. OOP medical expenses were commonly assessed on monthly,[31,35,43] quarterly[37,38,40] or annual[18-21,23,27,42] bases. Bourke-Taylor et al.[29] and Cohn et al.[28] reported expenses since diagnosis. In the case of OOP non-medical expenses, whereas five studies reported monthly figures,[31,35,36,39,43] others used shorter or longer assessment periods, ranging from daily[30] to annual[27,42] or even multi-year assessments.[28]

Seventeen studies aggregated individual indicators to estimate total OOP expenses.[17,22,23,27-32,35-40,42,43,46] Along with the numbers of aggregated indicators, their definitions varied. To provide a structured overview, we grouped all extracted indicators into 16 categories (5 OOP medical and 11 OOP non-medical). In addition, four studies estimated total OOP medical expenses above and beyond health insurance premiums or reimbursed healthcare costs without aggregating single indicators.[18-21] All indicators are listed in Table 2.

In terms of OOP medical expenses, the costs of pharmacotherapy (i.e., direct charges, co-payments and deductibles for prescription and non-prescription medications) were most frequently recorded. Overall, 10 studies assessed pharmacotherapy costs,[23,27,28,31,35-38,40,42,43] independently of context-specific characteristics such as healthcare systems or LLCs. Other OOP medical expenses, for example, therapeutic and surgical procedure costs, were only assessed within specific contexts. For instance, in India, Ahuja et al.,[32] measured radiation therapy and surgery costs to families of children with cancer because those families lacked appropriate insurance coverage. In addition, in Australia, Bourke-Taylor et al.[29] focused their study on equipment and assistive technology expenses, as government funding and insurance covered only a limited range of those, leaving some families to make up the, often, substantial difference.

Moreover, the overall number of included indicators varied considerably across studies. Tsimicalis et al.[38] assessed expenses for 16 indicators aggregating a total of 74 breakdown items, including specific medications (e.g., antibiotics, antiemetics, or antipyretics). In comparison, Ahuja et al.[32] reported expenses for only seven indicators. Both studies assessed OOP expenses incurred by families of children diagnosed with cancer.

Common indicators assessed regarding OOP non-medical expenses were illness-related costs for travel and transport,[17,22,27,28,30-32,35-40,42,43,46] accommodation,[17,22,28,30-32,37-39,42,43,46] and

food.[17,27,30-32,36-39,42,43,46] As with other matters, the degree of detail in which these costs were assessed and reported varied substantially across studies. Measuring travel expenses, Tsimicalis et al.[38] aggregated 11 breakdown items including taxi fares, public transit, parking and airfare. Other studies measuring OOP non-medical expenses provided total travel and transport expenses only, with no itemisation.[17,31,46]

Table 2: Categorised out-of-pocket expenses indicators.

Out-of-pocket medical expenses	Out-of-pocket non-medical expenses
Treatment and diagnostics 11/16 (69%)[a] - Pharmacotherapy[23,27,28,31,35-38,40,42,43] (prescription and non-prescription medications) - Therapeutic and surgical procedures[32] (e.g., surgery, radiation, transfusions) - Special treatments and supportive therapy[32,36-38,42,43] (e.g., chiropractor, massages, physiotherapy, rehabilitation services) - Complementary and alternative therapy[27,37,38,42] (e.g., homeopathy) - Investigations and diagnostic tests[31,32,36,42] (e.g., radiological investigations, laboratory tests) In-/outpatient charges and fees 7/16 (44%)[a] - Inpatient payments/hospital charges[23,31] - Public hospital bed charges[43] - Outpatient charges/payments[23,43] - Doctor/specialist fees[28,40,43] - Consultation fees[36,42] - Ward entrance fees[42] Equipment and assistive technology 3/16 (19%)[a] - Seating/standing equipment, specialised tables[29] - Specialised car, car modifications[29,37,38] - Mobility devices[29,36-38] (e.g., crutches, wheelchair) - Support equipment[29,37,38] (e.g., eye-wear, splints, monitoring devices, ventilator) - Communication devices[29] (e.g., displays, software) - Equipment for eating/drinking[29] (e.g., adapted cups, spoons, bottles, tubes) - Equipment for toileting, bathing and dressing[29] - Equipment for sleeping[29] (e.g., special mattress, body position support, height adjusted bed)	Travel and transport 15/16 (94%)[b] - Travel and transport[17,22,27,28,30-32,35-40,42,43,46] (e.g., airfare, gasoline, vehicle maintenance and registration, taxi, parking, public transport, roadway tolls, car rental, mileage) Accommodation 11/16 (69%)[b] - Accommodation[17,22,28,30-32,37-39,42,43,46] (e.g., lodging, room/bed rent, rented house, hotel/motel, hospitality house provided by volunteer families, hospital sponsored housing) Food 11/16 (69%)[b] - Additional food[17,27,30-32,36-39,42,43,46] (e.g., eating out, snacks, special diet foods during home stay) Childcare and home help 10/16 (63%)[b] - Childcare[17,27-29,36-38,43,46] - Home/domestic help[17,28,37-39,42,43,46] Communication, internet and cable TV 8/16 (50%)[b] - Communication[28,30,31,37-40,43,46] (frequently recorded: mail, mobile phone, landline costs) - Internet/cable TV[39] Clothing 5/16 (31%)[b] - Clothing bought because of the child's condition[28,31,37,38,43,46] Gifts, treats and toys 4/16 (25%)[b] - Gifts/treats for child or other members of family[37,38,43,46] - Toys/recreational opportunities[29] Daily necessities and hygiene products 4/16 (25%)[b] - Elements for personal and household hygiene[39] - Incidental expenses, daily necessities, supplies (e.g., batteries, toiletries)[17,37,38,42]

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Out-of-pocket medical expenses	Out-of-pocket non-medical expenses
Medical aids, dressings and disposables 3/16 (19%)[a] - Medical aids and dressings[28,42] - Medical disposables (e.g. gloves, syringes)[31] Other - Medical fees and costs for other family members/caregivers as a result of a child's illness[36-38] Total expenses 4/16 (25%)[a] - Total out-of-pocket medical expenses above and beyond health insurance premiums or reimbursed healthcare costs[18-21]	Utilities 2/16 (13%)[b] - Electricity,[39,43] gas,[39] water[39] Relocation and home modifications 2/16 (13%)[b] - Moving,[37,38] renovations,[37,38] home modifications[29] Other - Special education[28,42] - Attention to visitors[39] - School and extracurricular activities[38] - Pet care[38] - Funeral expenses[43]

TV indicates television.
[a] Number of studies measuring out-of-pocket medical expenses reporting at least one indicator in the respective category.
[b] Number of studies measuring out-of-pocket non-medical expenses reporting at least one indicator in the respective category.

2.5.4 Indirect costs: income loss and opportunity costs

Indirect costs were assessed in 15 studies[17,24-28,30,33,34,37,40-45] of which 14 recorded parental income loss.[17,24-28,30,33,34,37,40,42-45] The opportunity costs of informal caregiving were assessed in two studies.[37,41] These valued informal caregiving at market wage rates,[37,41] but based their calculations on different source data. Tsimicalis et al.[37] valued caregiving time according to US National Census Wage data; Pagano et al.[41] used a per diem based on the regional gross domestic product.

Measures used to quantify income loss varied. Most studies (n=11) used self-report data,[17,24-28,30,37,40,42,43] for instance, by using survey items with ordinal response categories[24-26] or valuing estimated work hours lost in relation to each family's income category.[17] Dockerty et al.[43] estimated income loss by subtracting each family's Consumer Price Index (CPI) -adjusted after-tax income before diagnosis from the CPI-adjusted after-tax income before participating in the study. Three studies linked data from government-administered registries to assess parental income loss.[33,34,44,45] These studies reported both changes in income over time and income disparities between families of children with and without specific LLCs. Nevertheless, overall, details on income composition were rarely reported. Outcome measures and valuation approaches are presented in Table 3.

Table 3: Categorised indirect cost measures.

Income loss	Opportunity costs
Self-report income data 11/15 (73%)[a] - Estimation of income loss by valuing illness-related work disruptions,[17,24-28,30,37,40,42] e.g.: ▪ Cutting back on work hours/commitments ▪ Quitting a job ▪ Forgone overtime ▪ Unpaid leave ▪ Closing/suspending business - Estimation of income loss by subtracting parent-reported income prior to diagnosis from parent-reported income prior to study participation[43] Income data from registers 3/15 (20%)[a] - Estimation of income loss by analysing income changes over several years comparing families of children with and without a particular health condition,[33,34,44,45] including various sources of income: ▪ Total income (all sources of income) ▪ Income from work ▪ Several types of benefits (e.g., unemployment, sickness, childcare related)	National Census Wage 1/15 (7%)[a] - Informal caregiving time spent by both mothers and fathers valued using US National Census Wage data[37] Regional gross domestic product 1/15 (7%)[a] - Informal caregiving time spent by one of the parents valued by the per diem regional gross domestic product[41]

[a] Number of studies measuring indirect costs grouped in the respective category.

2.5.5 Financial support

Four of our 28 reviewed studies reported the financial support families received.[27,31,43,46] Financial support was measured either in general terms[27,43,46] or specifically in relation to treatment.[31] Sources of financial support included various governmental[27,31,43,46] and non-governmental[27,31,46] sources, such as carers' allowances, charity grants, support groups or fund-raising efforts. Table 4 provides an overview of the types and sources of financial support provided.

Table 4: Type and source of financial support.

Type	Source
Assistance with medical expenses 4/28 (14%)[a] - Assistance with treatment expenses and care costs[27,31,43,46] Assistance with non-medical expenses 3/28 (11%)[a] - Assistance with non-medical expenses[27,43,46] (e.g., transportation, accommodation)	Governmental 4/28 (14%)[a] - Financial support received/available from governmental sources,[27,31,43,46] (e.g., disability living allowance, carer's allowance) Non-governmental 3/28 (11%)[a] - Financial support received from non-governmental sources,[27,31,46] (e.g., charities, support groups, fund-raising efforts)

[a] Number of total studies reporting financial support within the respective category.

2.6 Discussion

We retrieved and reviewed 30 publications reporting on 28 studies measuring financial burden in families of children with LLCs. We found that financial measures used to quantify financial burden fit into three broad categories: direct costs (OOP medical and non-medical expenses), indirect costs (income loss, opportunity costs) and financial support. While most reviewed studies covered one or two of these categories, none comprehensively assessed all three. Reporting gaps were particularly broad regarding incoming financial support and the opportunity costs of informal caregiving.

In addition, studies showed little consistency or standardisation concerning applied cost indicators and outcome measures. This is particularly apparent in assessments of OOP medical and non-medical expenses, with most studies measuring costs related to treatment (especially medication) and diagnostics, childcare and home help, food, accommodation and travel and transport. Further cost categories, as presented in Table 2, were only infrequently assessed, suggesting that, in some studies, direct costs are underestimated. In contrast, in studies using self-report data to estimate work-related income loss by estimating the effects of employment disruptions, indirect costs might be overestimated because the mitigating effects of income substitutes (e.g., benefits and allowances) are insufficiently recognised. Overall, outcome measures, methodological approaches and the extent to which study results were reported varied considerably across studies. These findings support those of previous reviews.[9-11]

Given that no consensus exists regarding the standardisation of financial burden measurement, with many cost categories and outcome measures assessed inconsistently across studies, we propose a framework for measuring direct costs (Table 5) as well as general recommendations for advancing research on financial burden and enhancing cost-of-illness studies' methodological consistency.

2.6.1 Direct costs

Considering the pronounced variations in the extent to which OOP expenses were assessed across studies and the fact that we could not locate any standardised sets of established cost indicators, our results suggest that not all direct costs were consistently measured across studies. Previous reviews have reported similar findings, indicating substantial inconsistencies in identifying, measuring and quantifying OOP expenses.[10,11] Nevertheless, accurate estimations

of financial burden require a comprehensive assessment of these expenses and should aim to fully identify, measure and evaluate the costs an illness imposes on families.

Regarding both healthcare systems and health conditions, this review included studies conducted in various settings and contexts. Although our extensive analyses gave us a comprehensive overview of a large variety of cost indicators, it also explains some of the heterogeneity in observed OOP expenses. While a number of indicators (e.g., prescription and non-prescription medication) may be applicable across contexts and settings, specific LLCs and healthcare system characteristics may require context and setting-specific indicators. For instance, as equipment and assistive technology needs vary among LLCs, and insurance coverage varies between healthcare systems, families in some areas may face substantial direct charges for treatment and procedures covered elsewhere by formal payers.

Regarding OOP non-medical expenses, studies commonly assessed childcare and home help, food, accommodation and travel and transport expenses. These expenses have been reported to contribute substantially to families' financial burden. In Canada, Tsimicalis et al.[38] found that travel, food and domestic help were the highest-ranked contributors, representing respectively 56%, 18% and 9% of the total direct costs to families of children with cancer. Other studies reported that communication as well as renovation and home modification expenses were major drivers of financial burden.[28,38] However, across the reviewed studies, these costs were infrequently assessed, indicating that direct costs may be commonly underestimated.

To enable more consistent and standardised assessments of financial burden, we used this study's findings to generate a framework for measuring direct costs in families of children with LLCs (Table 5). Consisting of 16 defined cost categories, this provides a basis for identifying, assessing and reporting cost indicators, many of which should be chosen in consideration of their relevance to the study's context and setting. Conducting and reporting cost-of-illness studies within this standardised framework will allow comprehensive, comparable assessments of OOP expenses and facilitate between-study comparisons. Methodological issues potentially arising with this framework's application are discussed below. While the proposed framework was based on direct costs incurred by families of children with LLCs, future research should consider its applicability to other chronic, but non-life-limiting, conditions.

Table 5: Framework for measuring direct costs among families of children with LLCs.

Direct Costs	Cost Categories	Definition	Further Considerations
Out-of-pocket medical	Treatment and diagnostics[a]	Treatment and diagnostics includes all family-incurred expenses related to procedures to diagnose and treat a disease or injury and improve health and health-related well-being.	- Out-of-pocket medical expenses include direct charges, co-payments, deductibles and co-insurances - Out-of-pocket non-medical expenses include additional illness related expenses above and beyond ordinary family-incurred costs - Sets of indicators for measuring out-of-pocket medical and non-medical expenses should be chosen in consideration of context-specific factors (e.g., healthcare system, health condition, phases of illness and treatment, different means of transportation) - Indicators should be defined in detail and their composition clearly stated - Further categories and indicators may apply - Inconsistencies in methodological approaches and biases associated with recall periods, data collection modes and aggregation issues should be rigorously addressed (see methodological considerations) - Consideration should be given to the research burden affected families might experience
	In-/outpatient charges and fees[a]	In-/outpatient charges and fees includes all family-incurred charges and fees for in- and outpatient services (e.g., hospital bed charges, consultation fees).	
	Equipment and assistive technology[a]	Equipment and assistive technology includes all family-incurred costs related to equipment and supportive technology needs.	
	Medical aids, dressings and disposables	Medical aids, dressings and disposables includes all occasional and incidental expenses that may be additionally incurred by families (e.g., gloves, wound dressings).	
	Other expenses	Other expenses includes potential further family-incurred out-of-pocket medical expenses (e.g., medical costs for other family members as a result of a child's illness).	
Out-of-pocket non-medical	Travel and transport	Travel and transport includes all family-incurred illness-related travel and transport expenses.[b]	
	Accommodation	Accommodation includes all family-incurred illness-related accommodation expenses.[b]	
	Food	Food includes all family-incurred illness-related expenses for food above and beyond ordinary expenses.[b]	
	Childcare and home help	Childcare and home help includes all family-incurred illness-related childcare and home help service expenses.	
	Communication, internet and cable TV	Communication, internet and cable TV includes all family-incurred illness-related expenses for communication, internet and cable TV above and beyond ordinary expenses.	
	Clothing	Clothing includes all family-incurred illness-related expenses for clothing above and beyond ordinary expenses.	

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Direct Costs	Cost Categories	Definition	Further Considerations
	Gifts, treats and toys	Gifts, treats and toys includes all family-incurred illness-related expenses for gifts, treats, toys and recreational opportunities above and beyond ordinary expenses.	
	Daily necessities and hygiene products	Daily necessities and hygiene products includes all occasional and incidental expenses that are additionally incurred by families because of a child's illness (e.g., batteries, soap).	
	Utilities	Utilities includes all family-incurred illness-related expenses for utilities (e.g., water, electricity, gas) above and beyond ordinary expenses.	
	Relocation and home modifications	Relocation and home modifications includes all family-incurred expenses for relocating or carrying out renovations and home modifications because of a child's illness.	
	Other expenses[a]	Other expenses includes all other family-incurred illness-related out-of-pocket non-medical expenses not captured by other categories.	

LLC indicates life-limiting condition; TV, television.
[a] Potential indicators, see Table 2.
[b] Potential cost items, see Table 2.

2.6.2 Indirect costs

Only half of the 28 included studies provided evidence on illness-related changes in parental income. Considering that a recent review on the impact of childhood cancer on parents' socio-economic situations observed a high prevalence of adverse employment effects, particularly among mothers,[9] attention to employment and income effects across reviewed studies seems rather low. One possible explanation is that no appropriate and reliable measurement tools are available to assess employment disruptions and income loss. Although most studies used self-reported employment disruption data to estimate work-related income loss, the reporting and valuation of these disruptions were highly inconsistent. For instance, although some studies recorded and valued specific employment disruptions (e.g., number of reduced working hours, days of unpaid leave), others used general estimates of overall income loss (e.g., by using ordinal response categories). We also noted that most studies disregarded the potential mitigating effects of financial coping strategies (e.g., income substitutions), thus likely overestimating income losses.[47,48]

Register-based studies more comprehensively assessed changes in parental income. As our findings showed, these studies not only examined income changes from a long-term perspective, but also included other types of income (e.g., unemployment and sickness benefits, fund raising) potentially alleviating income loss. Previous research indicates that income loss is most evident shortly after diagnosis.[24,40] From a long-term perspective, this suggests that transfer payments and coping strategies may mitigate these costs. Therefore, future studies should consider analysing long-term illness-related effects on parental income beyond employment disruptions.

Regarding the opportunity costs of informal caregiving, traditional assessments involve two steps: measuring time spent on it and valuing that time monetarily.[49] Depending on the chosen data collection mode, estimations may differ. In a comparison of diaries and surveys, diaries were found to reduce the risk of overestimations associated with long recall periods.[49] Moreover, opportunity costs may vary depending on the wage rate chosen to value caregiving time. Different valuation methods have been proposed, including the market wage rate, proxy wage rate and willingness to pay method.[50,51] Depending on the wage rate, then, opportunity costs may be over- or underestimated.[50]

2.6.3 Financial support

Only four studies included assessments of financial support, indicating a need for further research. Given that financial support (similar to the income substitutes discussed above) may mitigate the effects of illness-related costs, an assessment is critical to financial burden measurement. We observed little agreement regarding the reporting of this variable. Our experience supports findings by Roser et al.[9] In their systematic review, because different types and extents of financial support were reported, they had difficulty synthesising it.[9] Categorising financial support according to designated use (e.g., assistance with OOP medical or non-medical expenses) and source may increase reporting transparency.

2.6.4 Considerations regarding methodological approaches

Our inclusion of both longitudinal and cross-sectional studies explains some of the heterogeneity regarding reviewed studies' methodological approaches. Nevertheless, even in studies with similar designs, we found little consensus in terms of recall periods, data collection modes or data aggregation. Previous studies noted that these methodological inconsistencies constitute a major challenge for syntheses of research findings and cross-study comparisons.[9-11] For instance, variations in recall periods, with longer periods increasing the risk of recall errors,[52,53] render the accuracy of cost estimates somewhat uncertain. Therefore, shorter recall periods (e.g., 1 month) yielding more precise cost estimates are recommended.[54]

Additionally, cost estimates may vary depending on the data collection mode employed. Comparing diaries and surveys, for example, it has been suggested that cost diaries are more suitable for smaller, more frequent expenses, reducing recall errors, whereas surveys are thought to be reliable for capturing extraordinary, infrequent expenses.[52] Nevertheless, for families living with a child suffering from an LLC, keeping a cost diary could represent an additional burden during an already difficult time. Tsimicalis et al.[37,38] kept data collection periods short by combining interview surveys with one 1-week cost diary a month for three consecutive months. Validation of self-report data, for example, against official tax records (income loss) or receipts (OOP expenses), should also be considered to address recall errors.

We observed high variability both in the number of cost items (e.g., gasoline, parking tickets, road tolls) aggregated within an indicator (e.g., transportation) and in the overall number of included indicators aggregated to determine total OOP medical and non-medical expenses.

Previous research suggests that aggregated totals increase with increases in the number of measured items or indicators, increasing estimates' accuracy and reliability.[52,54] Therefore, using multiple indicators and cost items (as proposed in Table 5) might provide more precise estimates of financial burden.

2.6.5 Strengths and limitations

Using a scoping review methodology, we identified a wide variety of articles on illness-related costs incurred by families of children with LLCs. The scoping review methodology allowed us to include studies of various designs, thereby providing an ideal basis for mapping, consolidating and disseminating evidence on existing cost indicators and outcome measures. However, we neither analysed nor synthesised the extent to which specific expenses or employment disruptions contribute to families' financial burden. This limited our conclusion about their true monetary impact. Determining the relevance of each indicator and outcome measure within specific contexts and settings will require further research.

2.7 Conclusions

In addition to the physical and emotional challenges of caring for a child with an LLC, parents of these children face serious ongoing financial consequences. To facilitate evidence-based policy interventions that reduce the burden of illness-related costs, consistent and precise estimates of financial burden are necessary. Based on our findings, we recommend that future cost-of-illness studies cover all three relevant financial categories: direct costs, indirect costs and financial support. To help standardise this task, we generated a framework for measuring OOP expenses providing guidance for quantifying the full direct costs borne by families of children with LLCs. Moreover, overcoming inconsistencies regarding outcome measures and methodological approaches including recall periods, data collection modes and data aggregation methods, will strengthen inter-study comparisons and promote the development and implementation of effective support policies.

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2.9 Supplemental materials

Supplemental Table 1: Search strategy/string.

Medline, CINAHL, EconLit (via EBSCOhost)		Scopus	
No.	**Search Terms / Major Headings (MH)**	**No.**	**Search Terms**
1	TI children OR AB children	1	TITLE-ABS (children)
2	TI child OR AB child	2	TITLE-ABS (child)
3	TI childhood OR AB childhood	3	TITLE-ABS (childhood)
4	TI pediatric OR AB pediatric	4	TITLE-ABS (pediatric)
5	TI pediatrics OR AB pediatrics	5	TITLE-ABS (pediatrics)
6	TI paediatric OR AB paediatric	6	TITLE-ABS (adolescent)
7	TI paediatrics OR AB paediatrics	7	TITLE-ABS (infant)
8	TI adolescent OR AB adolescent	8	TITLE-ABS (minor)
9	TI adolescents OR AB adolescents		
10	TI infant OR AB infant		
11	TI infants OR AB infants		
12	TI minor OR AB minor		
13	TI minors OR AB minors		
14	MH “child”		
15	MH “pediatrics”		
16	MH “adolescent”		
17	MH “infant”		
18	MH “minors”		
19	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18	9	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8
20	TI family OR AB family	10	TITLE-ABS (family)
21	TI families OR AB families	11	TITLE-ABS (parent)
22	TI parents OR AB parents	12	TITLE-ABS (parental)
23	TI parent OR AB parent	13	TITLE-ABS (parenthood)
24	TI parental OR AB parental	14	TITLE-ABS (household)
25	TI parenthood OR AB parenthood	15	TITLE-ABS (caregiver)
26	TI household OR AB household	16	TITLE-ABS (“care-giver”)
27	TI households OR AB households		
28	TI caregiver OR AB caregiver		

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Medline, CINAHL, EconLit (via EBSCOhost)		Scopus	
No.	**Search Terms / Major Headings (MH)**	**No.**	**Search Terms**
29	TI caregivers OR AB caregivers		
30	TI “care-giver” OR AB “care-giver”		
31	TI “care-givers” OR AB “care-givers”		
32	MH “family”		
33	MH “parents”		
34	MH “home nursing economics”		
35	MH “caregivers economics”		
36	20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35	17	10 or 11 or 12 or 13 or 14 or 15 or 16
37	TI “economic burden” OR AB “economic burden”	18	TITLE-ABS (“economic burden”)
38	TI “financial burden” OR AB “financial burden”	19	TITLE-ABS (“financial burden”)
39	TI financial OR AB financial	20	TITLE-ABS (financial)
40	TI financials OR AB financials	21	TITLE-ABS (finance)
41	TI finance OR AB finance	22	TITLE-ABS (economic)
42	TI finances OR AB finances	23	TITLE-ABS (economics)
43	TI economic OR AB economic	24	TITLE-ABS (economical)
44	TI economics OR AB economics	25	TITLE-ABS (income)
45	TI economical OR AB economical	26	TITLE-ABS (salary)
46	TI income OR AB income	27	TITLE-ABS (wage)
47	TI salary OR AB salary	28	TITLE-ABS (“out-of-pocket”)
48	TI salaries OR AB salaries	29	TITLE-ABS (expenses)
49	TI wage OR AB wage	30	TITLE-ABS (expenditure)
50	TI wages OR AB wages	31	TITLE-ABS (cost)
51	TI “out-of-pocket” OR AB “out-of-pocket”		
52	TI expenses OR AB expenses		
53	TI expenditure OR AB expenditure		
54	TI expenditures OR AB expenditures		
55	TI cost OR AB cost		
56	TI costs OR AB costs		
57	MH “socioeconomic factors”		
58	MH “financing, personal”		
59	MH “economics”		
60	MH “income”		
61	MH “health expenditures”		

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Medline, CINAHL, EconLit (via EBSCOhost)		Scopus	
No.	**Search Terms / Major Headings (MH)**	**No.**	**Search Terms**
62	MH “cost of illness”		
63	MH “health care costs”		
64	37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 or 58 or 59 or 60 or 61 or 62 or 63	32	18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31
65	TI “special healthcare needs” OR AB “special healthcare needs”	33	TITLE-ABS (“special healthcare needs”)
66	TI “special health needs” OR AB “special health needs”	34	TITLE-ABS (“special health needs”)
67	TI “special health care needs” OR AB “special health care needs”	35	TITLE-ABS (“special health care needs”)
68	TI “special care needs” OR AB “special care needs”	36	TITLE-ABS (“special care needs”)
69	TI “complex health care needs” OR AB “complex health care needs”	37	TITLE-ABS (“complex health care needs”)
70	TI “complex healthcare needs” OR AB “complex healthcare needs”	38	TITLE-ABS (“complex healthcare needs”)
71	TI “complex health needs” OR AB “complex health needs”	39	TITLE-ABS (“complex health needs”)
72	TI “complex care needs” OR AB “complex care needs”	40	TITLE-ABS (“complex care needs”)
73	TI “special medical needs” OR AB “special medical needs”	41	TITLE-ABS (“special medical needs”)
74	TI “complex medical needs” OR AB “complex medical needs”	42	TITLE-ABS (“complex medical needs”)
75	TI “pediatric palliative care” OR AB “pediatric palliative care”	43	TITLE-ABS (“pediatric palliative care”)
76	TI “paediatric palliative care” OR AB “paediatric palliative care”	44	TITLE-ABS (“complex condition”)
77	TI “complex condition” OR AB “complex condition”	45	TITLE-ABS (“complex chronic condition”)
78	TI “complex conditions” OR AB “complex conditions”	46	TITLE-ABS (“complex medical condition”)
79	TI “complex chronic condition” OR AB “complex chronic condition”	47	TITLE-ABS (“life-limiting condition”)
80	TI “complex chronic conditions” OR AB “complex chronic conditions”	48	TITLE-ABS (“chronic health condition”)
81	TI “complex medical condition” OR AB “complex medical condition”	49	TITLE-ABS (cancer)
82	TI “complex medical conditions” OR AB “complex medical conditions”	50	TITLE-ABS (“nervous system disease”)
83	TI “life-limiting condition” OR AB “life-limiting condition”	51	TITLE-ABS (“cerebral palsy”)
84	TI “life-limiting conditions” OR AB “life-limiting conditions”	52	TITLE-ABS (“congenital heart disease”)
85	TI “chronic health condition” OR AB “chronic health condition”	53	TITLE-ABS (“chronic respiratory disease”)
86	TI “chronic health conditions” OR AB “chronic health conditions”	54	TITLE-ABS (“metabolic disorder”)
87	TI cancer OR AB cancer	55	TITLE-ABS (“congenital anomaly”)
88	TI “nervous system disease” OR AB “nervous system disease”		
89	TI “nervous system diseases” OR AB “nervous system diseases”		
90	TI “cerebral palsy” OR AB “cerebral palsy”		
91	TI “congenital heart disease” OR AB “congenital heart disease”		
92	TI “congenital heart diseases” OR AB “congenital heart diseases”		
93	TI “chronic respiratory disease” OR AB “chronic respiratory disease”		

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Medline, CINAHL, EconLit (via EBSCOhost)		Scopus	
No.	**Search Terms / Major Headings (MH)**	**No.**	**Search Terms**
94	TI “chronic respiratory diseases” OR AB “chronic respiratory diseases”		
95	TI “metabolic disorder” OR AB “metabolic disorder”		
96	TI “metabolic disorders” OR AB “metabolic disorders”		
97	TI “congenital anomaly” OR AB “congenital anomaly”		
98	TI “congenital anomalies” OR AB “congenital anomalies”		
99	MH “disabled children”		
100	MH “palliative care economics”		
101	MH “chronic disease economics”		
102	MH “neoplasms economics”		
103	MH “nervous system diseases economics”		
104	MH “heart diseases economics”		
105	MH “metabolic diseases economics”		
106	65 or 66 or 67 or 68 or 69 or 70 or 71 or 72 or 73 or 74 or 75 or 76 or 77 or 78 or 79 or 80 or 81 or 82 or 83 or 84 or 85 or 86 or 87 or 88 or 89 or 90 or 91 or 92 or 93 or 94 or 95 or 96 or 97 or 98 or 99 or 100 or 101 or 102 or 103 or 104 or 105	56	33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55
107	Final Search String: 19 AND 36 AND 64 AND 106	57	Final Search String: 9 AND 17 AND 32 AND 56

Chapter 3: Parenting a Child With a Life-Limiting Condition: Financial Impact of Hospitalisations and Death

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3.1 Highlights

- Families of children with life-limiting conditions are at risk of substantial financial burden from high out-of-pocket medical expenses. It is also known that parents often have to incur out-of-pocket non-medical expenses and reduce their work commitments.
- We provide new longitudinal evidence on the hospitalisation- and death-related financial and employment implications for families of children with life-limiting conditions. Child hospitalisations add to families' financial burden through increased travel and accommodation expenses. Work commitments rose during early bereavement.

3.2 Abstract

Objectives

To investigate out-of-pocket non-medical expenses and employment-related outcomes in families of children with life-limiting conditions. Specifically, to quantify the financial and employment implications of two events: a child's hospitalisation and death.

Methods

This cohort study used panel data collected prospectively for a larger study investigating the effectiveness of specialised paediatric palliative care. Participants were recruited by medical professionals between November 2019 and May 2022 at four Swiss children's hospitals. The care follow-up and bereavement follow-up assessments were 330 and 300 days, respectively. We measured out-of-pocket non-medical expenses, individual full-time equivalent units, personal income, as well as sick leave and vacation days taken. Analyses included descriptive statistics and two-way linear fixed-effects regressions.

Results

The analysis included 110 parents (mothers n=59, fathers n=51) of 61 children. Children were hospitalised for a median of 7 days (IQR 0–21, Rng. 0–227). The fixed-effects models found a positive association between hospitalisation, i.e., length of stay, and travel and accommodation expenses (Coef. 4.18, 95% CI 2.20–6.16). On average, for each week of hospitalisation, parents spent an additional CHF29 on travel and accommodation. During the 300-day bereavement follow-up, six (26%) of 23 parents increased their work commitments, while one reported a decrease.

Conclusions

Families incur higher travel and accommodation expenses during hospitalisation than during non-hospitalised periods. Instrumental support, e.g., hospital parking and food vouchers, can help families minimise these costs. Future studies should investigate whether early return to work during bereavement is driven by economic considerations or a desire for distraction.

3.3 Introduction

Parenting a child with a life-limiting condition (LLC), i.e., conditions '*for which there is no reasonable hope of cure and from which children will die*'[1,p.10] (Table 1), has an enormous impact on the life of these children's parents, adversely affecting their physical, psychological, emotional and social health.[2-6] In addition, these parents are likely to experience substantial economic adversity.[7] Previous research suggests that financial burden is driven by high out-of-pocket (OOP) medical spending, i.e., deductibles and co-payments.[8-10] However, the costs of taking care of a child with an LLC go beyond medical costs: parents often face OOP non-medical expenses and employment-related income losses.[11-13]

Over the course of an LLC, parents have to navigate a range of challenging and traumatic events, including unstable phases of illness, hospital admissions and even their child's death, any of which can increase their psychological, emotional, social and financial burdens.[2-6,14-17] For example, families may face extra expenses for travelling to and staying overnight at the hospital. Repeated and lengthy hospitalisations are common for children with LLCs.[18,19] However, few studies have examined the effects of a child's hospitalisation on OOP non-medical expenses and parental employment.[9,20-24] None of these studies is both LLC-specific and longitudinal.[9,20-24]

The same is true for evidence on the financial and employment implications of a child's death. Losing a child is a traumatic, life-changing event.[25-29] Conducting research during early bereavement is challenging, as parents are navigating an unimaginably difficult period of their lives.[30] As a result, studies about the economic consequences of grief are largely absent from the literature. Nevertheless, the financial and employment implications of losing a child to an LLC can be considerable.[31]

For this study, we investigated the financial and employment implications of both hospitalisation and bereavement. More specifically, the study's first aim was to explore how a child's

hospitalisation influences the family's OOP non-medical expenses as well as the parents' income and employment over a 330-day period. Its second aim was to assess changes in parental income and employment over the first 300 days of bereavement.

Table 1: Categories of LLCs in children[a].

Category	LLCs in Children
1	Conditions for which curative treatment may be feasible but can fail (e.g., cancer, irreversible organ failures)
2	Conditions in which premature death is inevitable but long periods of intensive treatment are likely (e.g., Duchenne muscular dystrophy)
3	Progressive conditions without curative treatment options (e.g., severe metabolic conditions)
4	Irreversible but nonprogressive conditions that cause severe disability (e.g., severe cerebral palsy)

[a] Categories of LLCs in children as defined by Together for Short Lives (2018).

3.4 Methods

This cohort study used a prospectively collected panel dataset to explore the financial and employment implications of childhood hospitalisation and death in families of children with an LLC. The dataset was collected through the 'Specialised Paediatric PAlliativE CaRe: Assessing family, healthcare professional and health system outcomes in a multi-site context of various care settings (SPhAERA)' study, which aimed to evaluate the effectiveness of a Swiss specialised paediatric palliative care (SPPC) programme.[32] The study was conducted between November 2019 and May 2023 with the approval of the responsible Swiss ethics committees (BASEC-Nr. 2019–01170) and is registered in ClinicalTrials.gov (NCT04236180). Further information regarding the SPhAERA study can be found elsewhere.[32]

3.4.1 Participants and recruitment

This study's population consisted of children with an LLC and their parents (mothers and/or fathers). To be included for study participation, children had to be 0–18 years of age and their families had to be proficient in either German or French. Children whose life expectancy was <48 hours and neonates with medical complications due to prematurity and/or birth complications treated in a neonatal intensive care unit were excluded. Children subject to child protection regulations and their parents were ineligible for study participation.

Participants were recruited between November 2019 and May 2022 at four Swiss study centres, i.e., four (University) Children's Hospitals. In one study centre, participants were recruited consecutively when they entered the local SPPC programme. In the other three study centres, participants potentially in need of SPPC were convenience recruited by the responsible medical professional. Parents provided written informed consent.

3.4.2 Data collection and variables

Data collection

The study duration was 330 days, with the exception that, in the event of a child's death, parents remained enrolled for an additional 300 days, starting from the date of death. Diagnostic information and healthcare resource utilisation data were extracted from routine data via chart reviews at baseline (day 0) and at eight care follow-up assessments, i.e., at days 15, 30, 60, 90, 120, 150, 240 and 330. Healthcare resource utilisation data collected at days 15 and 30 were merged to be in line with the collection of economic data. The collection of economic data started at day 30 and continued throughout days 60, 90, 120, 150, 240 and 330. During the bereavement follow-up, economic data were collected at days 120 and 300. Parent and family characteristics and economic data were collected via paper-pencil self-report questionnaires.

Exposure variable

To explore associations between hospitalisation and family economic outcomes, we used child hospital length of stay (LoS, days) as our exposure measure. For each assessment period, the number of days a child was hospitalised was recorded.

Outcome variables

In addition to a range of OOP non-medical expenses, outcome variables included individual full-time equivalent (FTE) units, personal income and work absences, i.e., sick leave and vacation days. While work absenteeism may not have immediate financial implications, it potentially hampers parents' long-term career perspectives.[33] Families' OOP non-medical expenses included illness-related expenses for home healthcare supplies, travel and accommodation, childcare and home help, and special and extraordinary purchases, e.g., home modifications. To measure work commitment and income loss, each parent's FTE unit and income were recorded at study start. At each care follow-up assessment, parents who had experienced work

and/or income-related changes were asked to provide their new FTE unit and/or income. Work absences (both paid and unpaid) of employed parents included two variables: sick leave days and vacation days. In the bereavement follow-up, individual FTE units, income and work absenteeism were measured. All expenses and income were measured in Swiss francs (CHF).

Other variables

We collected data on a number of relevant diagnostic and socio-demographic/economic characteristics. We also assessed the number of hospitalisations and the amount of financial support families received.

3.4.3 Statistical analyses

Descriptive statistics were used to provide an overview of parent, family and child characteristics, to report on the financial support families received and to describe financial and employment implications. Missing data were analysed using Little's test.[34]

To estimate the associations of child hospital LoS with our outcome variables, we used two-way linear fixed-effects models. The empirical model is specified in Supplemental Material 1. By including subject- and time-fixed effects, fixed-effects models implicitly control for variables that are constant over time but differ across subjects, as well as for those variables that are constant across subjects but change over time.[35,36] This controls for any time-invariant factors that may have confounding effects or increase the data's heterogeneity.[35,36] As financial support varies with time and between subjects, we included this variable in our adjusted models. In addition, a dichotomous indicator for period length, i.e., short (30 days) or long (90 days), was added to account for the differences in observation length. In the parent-level analyses, a child-fixed effect was included to account for within-family correlations. For all analyses, cluster-robust standard errors were used. For the regression analyses, categorical outcome variables were converted to continuous variables by assigning the midpoint of each categorical range as the representative value and using a conservative estimate of the categories' lower limit +1 for open-ended categories. In presenting the regression outputs, our reference group—consisting of all children who did not have any hospital stay during study participation—functioned as a benchmark. All analyses were performed using the R statistical software (version 4.1.2).[37] The regression analyses were performed using the R plm package (version 2.6-2).[38] A p-value of <0.05 was applied.

Robustness check

We checked the robustness of our findings via random-effects, complete-case and subgroup analyses. Fixed- and random-effects models were compared using the Hausman test (Supplemental Table 1).[39] Although both the fixed-effects and the random-effects models were consistent, we chose to report the former because of their ability to control for time-invariant unobserved heterogeneity and omitted variables. The results of the random-effects models are presented in Supplemental Table 2.

To check whether our results are robust regarding attrition and death, a binary variable indicating full 330-day study participation was created. To test for the possibility that limited exposure and high income could mitigate potential effects, we specified subgroups based respectively on total hospital LoS and household income. Also, because the association of hospital LoS with both home healthcare supply expenses and special and extraordinary expenses could vary by diagnosis, we created dichotomous diagnosis-specific subgroups, i.e., neurological vs. non-neurological. The association between hospital LoS and travel and accommodation expenses was tested based on home-to-hospital travel distance.

3.5 Results

3.5.1 Study participants

The inclusion criteria were met by 160 of 280 screened families. Of these 160 families, 70 consented for study participation. A family of twins was counted twice because both children participated in the study. Parents who did not complete an assessment at day 30 for reasons other than a child's death were excluded. Overall, the sample utilised in this study's analyses consisted of 110 parents of 61 children with LLCs (Figure 1).

Parent, family and child characteristics are presented in Table 2. Among parents with foreign citizenship, 12 (48%) were German. The majority of parents not in employment at study entry were mothers (n=13, 87%). Fifty-three families (87%) had supplementary insurance in addition to compulsory health insurance.

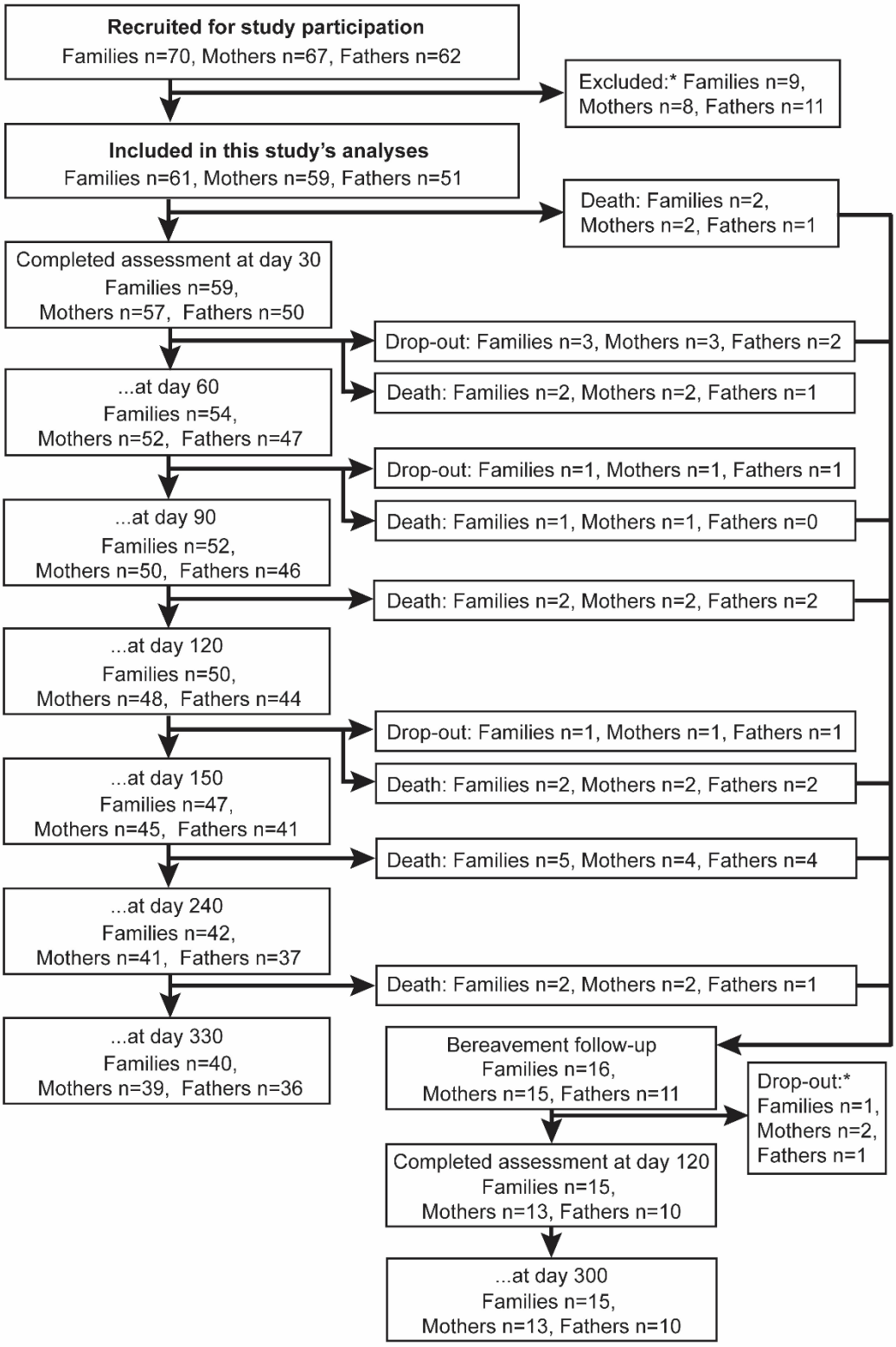

Figure 1: Flow diagram of study participants. The number of families is equal to the number of children; one family of twins was counted twice. Parents/families who did not complete assessments at day 30 or participate in the bereavement follow-up were excluded. *Only one parent (mother or father) was excluded/dropped-out.

Table 2: Parent, family and child characteristics.

Parents	**(n=110)**[a]
Parents, n (%)	
Mothers / Fathers	59 (54%) / 51 (46%)
Age in years,	
Mean (SD)	38.7 (6.5)
Nationality, n (%)	
Switzerland	85 (77%)
Other	25 (23%)
Time others have lived in Switzerland, n (%)	
>10 years	18 (72%)
<10 years	7 (28%)
Marital status, n (%)	
Married, Partnership	100 (91%)
Other	10 (9%)
Living situation, n (%)	
Couple family with child(ren)	96 (87%)
Other	14 (13%)
Number of children,[b]	
Median (IQR)	2.0 (1.0–2.0)
Range	1.0–5.0
missing, n (%)	*1 (1%)*
Education, n (%)	
Primary/secondary education or high school	8 (7%)
Vocational training	38 (35%)
College of higher education	36 (33%)
University degree	28 (25%)
Occupational status, n (%)	
In employment	95 (86%)
Not in employment	15 (14%)

Families	**(n=61)**[a]
Household income in CHF, n (%)	
<100'000	25 (41%)
100'000–200'000	26 (43%)
>200'000	8 (13%)
missing	*2 (3%)*
Home-hospital travel distance, n (%)	
0–20 kilometres	23 (38%)
21–50 kilometres	27 (44%)
>50 kilometres	11 (18%)

Children	**(n=61)**
Gender, n (%)	
Female / Male	34 (56%) / 27 (44%)
Age in years,	
Median (IQR)	3.6 (0.7–8.6)
Range	0.0–15.5
Diagnosis, n (%)	
Neurological	41 (67%)
Cardiological	10 (16%)
Oncological	6 (10%)
Other	4 (7%)
Illness duration in days,[c]	
Median (IQR)	357.0 (44.0–1827.0)
Range	4.0–5640.0
Place of care at study entry, n (%)	
Hospital	24 (39%)
Home	33 (54%)
Other	4 (7%)

SD indicates standard deviation; IQR, inter quartile range; CHF, Swiss francs.
[a] A family (two parents) of twins was counted twice because both children had an LLC and participated in the study.
[b] Number of children includes the child with an LLC.
[c] Illness duration gives the number of days between the date of diagnosis of the LLC and the date of study entry.

3.5.2 Hospitalisations and LoS

Over the full study period, the median number of hospitalisations per child was 1 (IQR 0–3, Rng. 0–6). For the 30-day and 90-day assessment periods, the maximum number of hospitalisations per child was 2 and 3, respectively. Seventeen children (28%) had no hospitalisation. The median total hospital LoS was 7 days (IQR 0–21, Rng. 0–227). Details on child hospital LoS are provided in Figure 2.

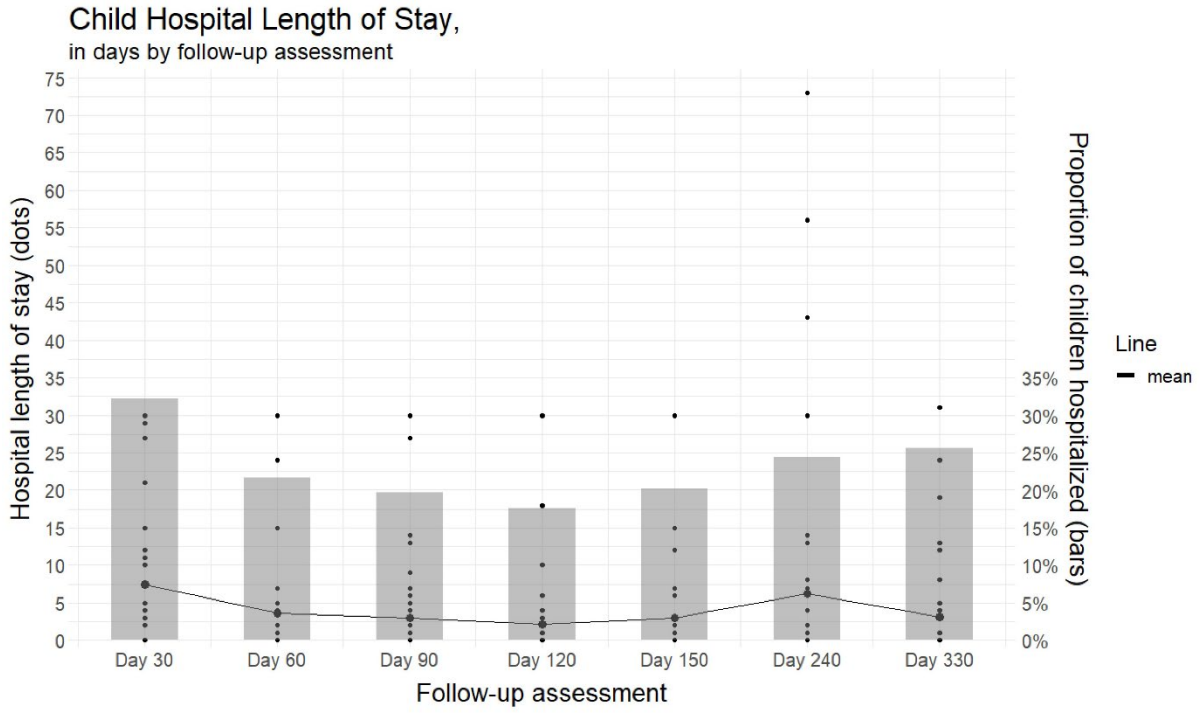

Figure 2: Child hospital LoS. Assessment periods were 30 days each for the first five assessments and 90 days each for the last two. The line gives the mean child hospital LoS. Each bar indicates the proportion of children hospitalised for at least one day for the corresponding assessment.

3.5.3 Economic outcomes

Figures 3A–D and 4A–E detail the outcomes of the 330-day follow-up. The proportion of families that did not incur any travel and accommodation expenses peaked at day 120—the same day the proportion of children hospitalised was lowest. Childcare and home help expenses averaged CHF480 per month for the 20 families (33%) that had such expenses. Throughout study participation, six families (10%) spent >CHF10’000 for special and extraordinary purchases. The highest share of parents (n=9, 15%) decreasing their individual FTE, primarily mothers (n=7, 78%), was observed on day 30. Throughout the 330-day follow-up, an average of 33%

of mothers were not in employment, while for fathers this applied to 5%. The amount of missing data was highest for sick leave and vacation days (up to 21%) and lowest for OOP non-medical expenses (5% max.) (Supplemental Table 3). As for the latter, Little's test showed that missing data were likely to be completely random (Supplemental Table 4). At each assessment, 50% or fewer families had financial support (Supplemental Table 5).

Sixteen children (26%) died during study participation. Deaths occurred at a median of 132 days (IQR 87–224, Rng. 6–252) after study entry. Locations of death were: hospital (n=8, 50%), home (n=7, 44%) and other (n=1, 6%). At the last follow-up assessment before death, five (19%) of the 26 participating parents of these 16 deceased children were not in employment (missing n=3, 12%). Of the 23 parents (88%) who completed the bereavement assessment on day 120, five (22%) had increased their work commitments, while one (4%) reported a decrease. During the first 120 days of bereavement, four parents (17%) took >30 days of sick leave, while two (9%) took >30 vacation days. Parents increasing their work commitments during bereavement also reported an increase in income (Supplemental Table 6).

3.5.4 Hospitalisation-related implications

The seven care follow-up assessments provided 344 family and 633 parent observations for use in the regression analyses. The adjusted fixed-effects analyses showed a positive association of child hospital LoS with travel and accommodation expenses (Coef. 4.18, 95% CI 2.20–6.16). For every one-day increase in hospital LoS, travel and accommodation expenses increased by 6.2% compared to the reference group. The median hospital LoS was 7 days (IQR 0–21, Rng. 0–227). With a mean difference in travel and accommodation expenses of CHF4.18, families spent an additional CHF29 for every week of their child's hospitalisation. No other associations were found between child hospital LoS and our other outcomes (Table 3).

3.5.5 Robustness check

The complete-case and subgroup analyses support the findings of our fixed-effects analyses, with two exceptions: Hospital LoS was no longer associated with travel and accommodation expenses for families with a household income of ≥CHF100'000 and those with a home-to-hospital travel distance of ≤20 kilometres (Supplemental Tables 7–12).

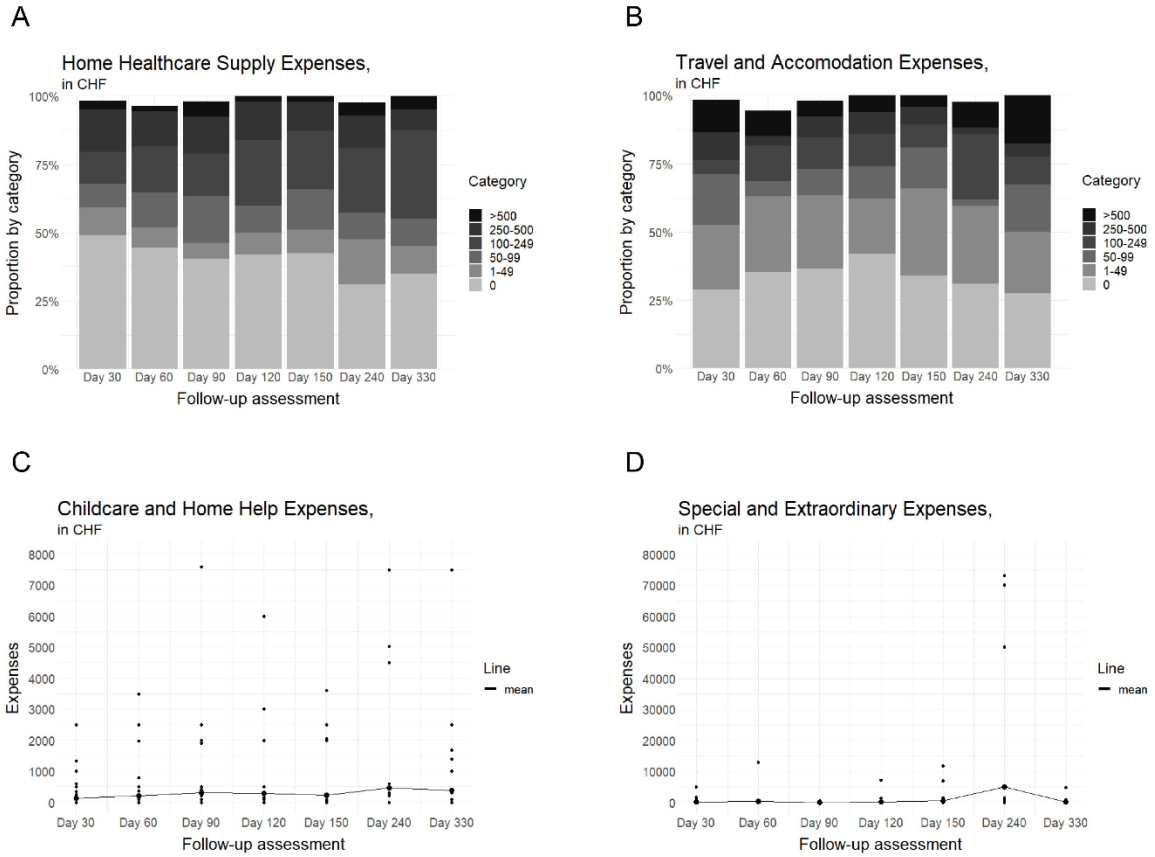

Figure 3A-D: Family OOP expenses. Assessment periods were 30 days each for the first five assessments and 90 days each for the last two. In Subfigures A and B, the sum of the proportions is less than 100% due to missing data.

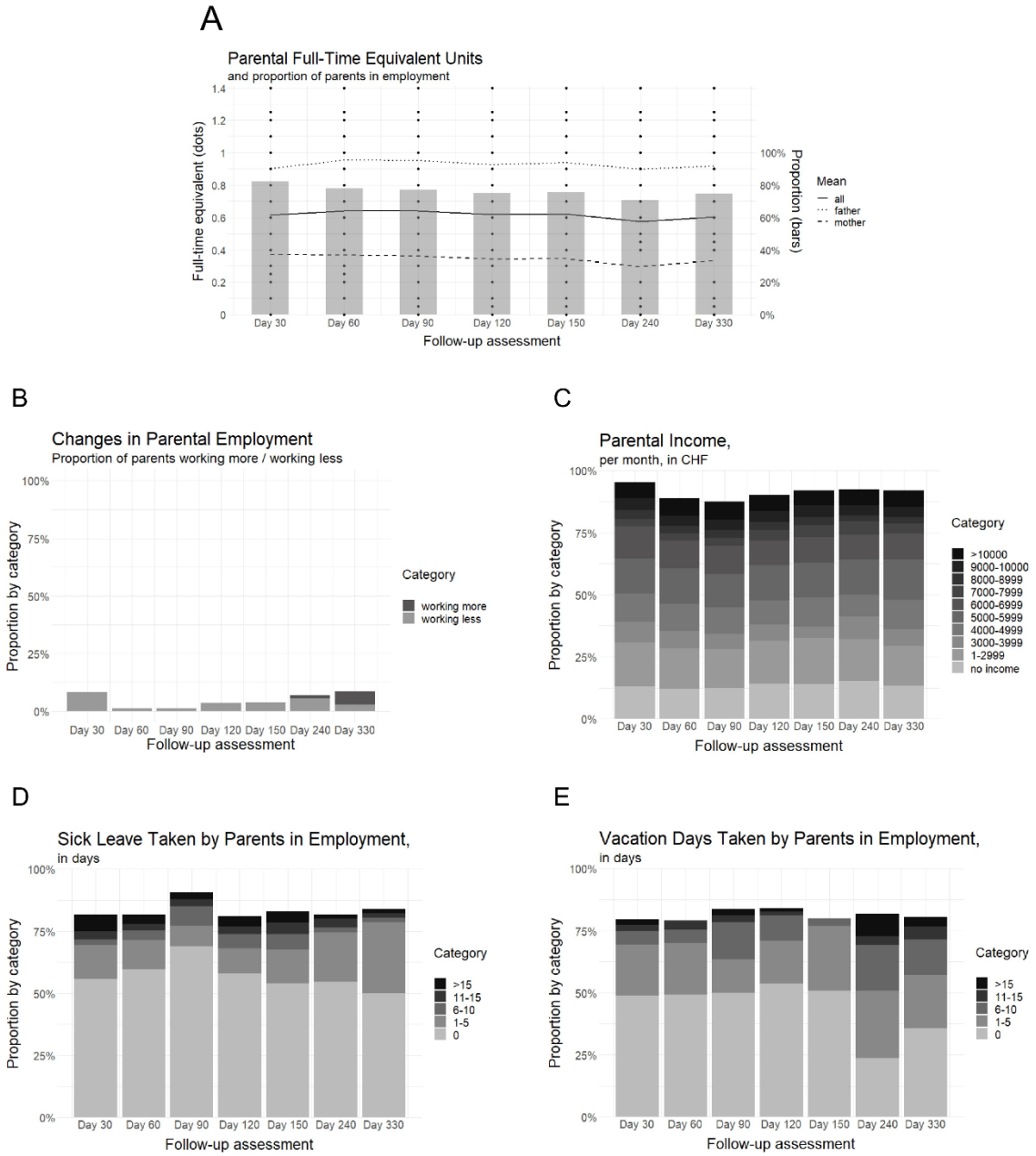

Figure 4A-E: Parental FTE, income and work absences. Assessment periods were 30 days each for the first five assessments and 90 days each for the last two. The sum of the proportions is less than 100% due to missing data. Some parents reported >1FTE, e.g., because of smaller side-line enterprises.

Table 3: Crude and adjusted two-way linear fixed-effects models of the effect of child hospital LoS on family/parent economic outcomes.

Outcome	Crude models[a]				Adjusted models[b]					
	Obser-vations[e]	Coef.	[95% CI]	*p*	Obser-vations[e]	Coef.	[95% CI]	*p*	Mean reference group[f]	Relative effect[g]
Out-of-pocket expenses										
Home health-care supplies[c]	339	-1.33	[-3.37 to 0.70]	*0.20*	329	-1.29	[-3.33 to 0.74]	*0.21*	181.50	-0.7%
Travel and ac-commodation[c]	338	4.46	[2.52 to 6.41]	*<0.001*	328	4.18	[2.20 to 6.16]	*<0.001*	67.76	6.2%
Childcare and home help[c]	339	-3.40	[-15.52 to 8.72]	*0.58*	329	-3.92	[-12.77 to 4.93]	*0.39*	542.33	-0.7%
Special and extraordinary[c]	338	-46.01	[-156.71 to 64.69]	*0.42*	328	-52.38	[-165.67 to 60.91]	*0.37*	1843.99	-2.8%
Employment and income										
Full-time equiv-alent unit	600	-0.00	[-0.00 to 0.00]	*0.99*	585	0.00	[-0.00 to 0.00]	*0.92*	0.60	0.0%
Income[c]	577	1.69	[-7.73 to 11.11]	*0.73*	564	2.90	[-6.74 to 12.54]	*0.56*	4822.83	0.1%
Work absen-teeism										
Sick leave[d]	404	0.03	[-0.02 to 0.08]	*0.30*	401	0.03	[-0.02 to 0.08]	*0.27*	2.02	1.5%
Vacation[d]	393	-0.06	[-0.13 to 0.01]	*0.10*	388	-0.06	[-0.14 to 0.01]	*0.08*	2.23	-2.7%

Coef. indicates coefficient; CI, confidence interval.
Note: Estimated coefficients represent mean differences in outcomes per day of hospitalisation.
[a] The crude models are linear fixed-effects models with time- and subject-fixed effects.
[b] The adjusted models are linear fixed-effects models with time- and subject-fixed effects adjusted for financial support and follow-up assessment period length.
[c] in Swiss francs.
[d] in days.
[e] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).
[f] The reference group used as a benchmark included all children who did not have any hospital stay during study participation.
[g] Relative effect = (estimated coefficient of the adjusted model / mean expenses reference group) x 100.

3.6 Discussion

In a sample of parents of children with an LLC, we investigated the financial and employment implications of two events: child hospitalisation and death. Child hospitalisation, i.e., hospital LoS, was positively associated with families' travel and accommodation expenses. During the first 120 days of bereavement, more than one-fifth of grieving parents increased their work commitments.

The additional travel and accommodation expenses endured by families are likely explained by parents maintaining a bedside presence during their child's hospitalisation, e.g., for transportation, parking, board and lodging.[20-22,24] In our sample, the extra travel and accommodation expenses incurred by parents per week of hospitalisation were rather low. For some parents, setting-specific factors such as parking and food vouchers or free-of-charge hospital accommodation for parents, may have limited these expenses. However, in settings with less support, these expenses could be much higher. Moreover, with repeated and lengthy hospitalisations, travel and accommodation expenses are likely to be greater.

Consistent with previous research, our analyses indicate that families residing farther away from the hospital experience increased travel and accommodation expenses.[20,22] Families with lower income may have reported their spending more accurately, as they were required to provide detailed records to financial assistance programmes. This may explain the association of hospital LoS with travel and accommodation expenses for these families. Another explanation could be that, due to affordable housing options, families with lower income live farther away from hospitals.

There are several possible explanations for the lack of association between hospital LoS and other OOP non-medical expenses. First, during their child's hospitalisation, a family may purchase home healthcare supplies in preparation for their child's hospital-to-home transition. Being prepared in terms of medication, equipment and supplies has been shown to be a priority for families.[40] Second, informal care provided by relatives and friends may explain both the low number of families that incurred formal childcare and home help expenses and the lack of association between such expenses and hospital LoS. Here, our findings contrast with previous research, which suggested that childcare expenses, e.g., for siblings, would normally increase during hospitalisations.[20,21] Third, a child's functional limitations, namely their dependency on medical technology and equipment, may be a stronger explanatory factor for increased special

and extraordinary expenses than hospitalisation.[12] Although few families in our study incurred such expenses, for those who did, they were exceedingly high.

Contrary to our expectations, we did not find any significant hospitalisation-related employment or income implications. Income replacement mechanisms, e.g., paid sick leave, and flexible employment arrangements, such as working from home, may have protected families from adverse effects. Previous research suggests that income loss is less severe in parents with flexible work arrangements.[21] As a contextual matter, it is worth noting that data collection occurred during the coronavirus disease 2019 pandemic, which brought a substantial increase in flexible work practices.[41]

In addition, families were recruited a median of one year after their child's diagnosis, when certain employment adaptations may already have taken place. For instance, mothers of children diagnosed during maternity leave may have extended that leave. A previous study of employment implications in families of children with special healthcare needs found that forgone employment was disproportionately high among mothers of young children (0–5 years).[42] Compared to mothers in the general population, those in our study were less likely to be employed. Across the care follow-up assessments, an average of 33% of participating mothers did not engage in employment, compared with 17% in the general population.[43]

Regarding the bereavement follow-up, parents' increasing work commitments may have been motived by both economic considerations and a desire to resume work, e.g., as a means of distraction.[31,44] I.e., the observed increases in work commitment do not necessarily indicate full readiness or restored functionality.[31] In general paediatrics, parental grief-related costs associated with work-presenteeism outweighed those associated with work-absenteeism.[31] The same principle may also explain a number of our bereavement follow-up observations of parental income, sick leave and vacation days.

Strengths and limitations

One of this study's major strengths is its longitudinal design: multiple assessments provided us with a realistic representation of how LLC-related events—and costs—develop over time. Another is its fixed-effects approach, which controls for time-invariant heterogeneity and confounders. Nevertheless, it also has notable limitations, including the relatively small sample size, which limits its statistical power and generalisability. In addition, sample bias cannot be ruled out, as families experiencing high psychological, emotional, social or financial burdens

may have been more likely to decline study participation. Moreover, outcome data were self-reported, leaving them vulnerable to response and recall bias. Some parents may have been reluctant to disclose their income. However, our instruments' discrete response categories and primarily monthly assessments would have minimised this type of bias. Finally, hospitalisation rates may have been influenced by the coronavirus disease 2019 pandemic.

3.7 Conclusions

Hospitalisation-related travel and accommodation expenses exacerbate the often substantial financial burden experienced by families of children with LLCs. To protect these families' financial well-being, adequate financial support should be made available to them. Where such financial support currently exists, additional efforts should be made both to enhance affected parents' awareness and assist them with the application processes. Future research is also needed to further explore the effect of LLCs in children on their parents' employment. To date, little is known of the factors behind these parents' decisions to reduce or increase their work commitments through their children's illness and their own bereavement.

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3.9 Supplemental materials

Supplemental Material 1: Empirical model.

Empirical model:

$$\boldsymbol{Y_{it} = \alpha_i + \beta_1 LoS_{it} + \ \ldots \ + \beta_k X_{k,it} + \lambda_t + \varepsilon_{it}}$$

Where **Y** represents the economic outcome of interest in a subject (i.e., family/parent) **i** at time **t**. The coefficient **β** can be interpreted as the within-subject effect on outcome Y of having been exposed to child hospital LoS ($\mathbf{LoS_{it}}$). The subject fixed-effects are given by $\boldsymbol{\alpha_i}$, representing the subject-specific intercepts (α_1, …, α_n). The time fixed-effects are given by $\boldsymbol{\lambda_t}$, representing the intercept of each time period. $\mathbf{X_{2,it}}$, …, $\mathbf{X_{k,it}}$ are other observed determinants of Y that are correlated with LoS_{it} and that vary across subjects and over time, e.g., financial support. $\boldsymbol{\varepsilon_{it}}$ is the unobserved error term.

Supplemental Table 1: Results of the Hausman test.

Outcome variable	Adjusted fixed-effects model vs. adjusted random-effects model
	p
Out-of-pocket expenses	
Home healthcare supplies	*0.30*[a]
Travel and accommodation	*0.94*[a]
Childcare and home help	*0.86*[a]
Special and extraordinary	*0.28*[a]
Employment and income	
Full-time equivalent unit	*0.09*[a]
Income	*0.32*[a]
Work absenteeism	
Sick leave days	*0.47*[a]
Vacation days	*0.06*[a]

[a] A p-value of >0.05 suggests that there is no significant evidence to reject the null hypothesis that the fixed-effects model is preferred, indicating that both models are consistent.

Supplemental Table 2: Crude and adjusted random-effects models of the effect of a child's hospital LoS on family/parent economic outcomes.

Outcome	Crude models[a]				Adjusted models[b]					
	Observations[e]	Coef.	[95% CI]	*p*	Observations[e]	Coef.	[95% CI]	*p*	Mean reference group[f]	Relative effect[g]
Out-of-pocket expenses										
Home healthcare supplies[c]	339	-2.07	[-3.73 to -0.42]	*0.01*	329	-2.10	[-3.73 to -0.47]	*0.01*	181.50	-1.2%
Travel and accommodation[c]	338	4.44	[2.75 to 6.13]	*<0.001*	328	4.21	[2.51 to 5.91]	*<0.001*	67.76	6.2%
Childcare and home help[c]	339	-4.55	[-14.93 to 5.84]	*0.39*	329	-4.92	[-12.51 to 2.68]	*0.20*	542.33	-0.9%
Special and extraordinary[c]	338	-25.02	[-93.24 to 43.20]	*0.47*	328	-26.28	[-94.62 to 42.06]	*0.45*	1843.99	-1.4%
Employment and income										
Full-time equivalent unit	600	-0.00	[-0.00 to 0.00]	*0.67*	585	0.00	[-0.00 to 0.00]	*0.93*	0.60	0.0%
Income[c]	577	1.39	[-7.67 to 10.45]	*0.76*	564	3.18	[-6.10 to 12.46]	*0.50*	4822.83	0.1%
Work absenteeism										
Sick leave[d]	404	0.03	[-0.01 to 0.08]	*0.16*	401	0.04	[-0.01 to 0.08]	*0.14*	2.02	2.0%
Vacation[d]	393	-0.00	[-0.06 to 0.05]	*0.95*	388	-0.01	[-0.06 to 0.04]	*0.76*	2.23	-0.4%

Coef. indicates coefficient; CI, confidence interval.
Note: Estimated coefficients represent mean differences in outcomes per day of hospitalisation.
[a] The crude models are random-effects models with time- and subject-fixed effects.
[b] The adjusted models are random-effects models with time- and subject-fixed effects adjusted for financial support and follow-up assessment period length.
[c] in Swiss francs.
[d] in days.
[e] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).
[f] The reference group used as a benchmark included all children who did not have any hospital stay during study participation.
[g] Relative effect = (estimated coefficient of the adjusted model / mean expenses reference group) x 100.

Supplemental Table 3: Outcome data of the 330-day care follow-up assessment.

Outcome	Day 30	Day 60	Day 90	Day 120	Day 150	Day 240	Day 330
Out-of-pocket expenses in CHF	**Families n=59**	**Families n=54**	**Families n=52**	**Families n=50**	**Families n=47**	**Families n=42**	**Families n=40**
Home healthcare supplies, n (%)							
0	29 (49%)	24 (44%)	21 (40%)	21 (42%)	20 (43%)	13 (31%)	14 (35%)
1–49	6 (10%)	4 (7%)	3 (6%)	4 (8%)	4 (8%)	7 (17%)	4 (10%)
50–99	5 (9%)	7 (13%)	9 (17%)	5 (10%)	7 (15%)	4 (9%)	4 (10%)
100–249	7 (12%)	9 (17%)	8 (15%)	12 (24%)	10 (21%)	10 (24%)	13 (33%)
250–500	9 (15%)	7 (13%)	7 (14%)	7 (14%)	5 (11%)	5 (12%)	3 (7%)
>500	2 (3%)	1 (2%)	3 (6%)	1 (2%)	1 (2%)	2 (5%)	2 (5%)
missing	*1 (2%)*	*2 (4%)*	*1 (2%)*	-	-	*1 (2%)*	-
Travel and accommodation, n (%)							
0	17 (29%)	19 (35%)	19 (37%)	21 (42%)	16 (34%)	13 (31%)	11 (27%)
1–49	14 (24%)	15 (28%)	14 (27%)	10 (20%)	15 (32%)	12 (29%)	9 (23%)
50–99	11 (18%)	3 (6%)	5 (10%)	6 (12%)	7 (15%)	1 (2%)	7 (18%)
100–249	3 (5%)	7 (13%)	6 (12%)	6 (12%)	4 (9%)	10 (24%)	4 (10%)
250–500	6 (10%)	2 (4%)	4 (8%)	4 (8%)	3 (6%)	1 (2%)	2 (5%)
>500	7 (12%)	5 (9%)	3 (6%)	3 (6%)	2 (4%)	4 (10%)	7 (18%)
missing	*1 (2%)*	*3 (6%)*	*1 (2%)*	-	-	*1 (2%)*	-
Childcare and home help							
Mean (SD)	137.1 (414.9)	199.8 (649.4)	307.8 (1’161.9)	289.0 (1’002.4)	223.2 (739.0)	459.5 (1’531.4)	387.3 (1’268.2)
Median (IQR)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–25)
Range	0–2’500	0–3’500	0–7’600	0–6’000	0–3’600	0–7’500	0–7’500
missing, no (%)	*1 (2%)*	*2 (4%)*	*1 (2%)*	-	-	*1 (2%)*	-
Special and extraordinary							
Mean (SD)	211.5 (725.7)	297.1 (1801.0)	23.9 (93.3)	178.8 (1’017.6)	464.7 (1’976.2)	4’903.9 (17’398.3)	197.5 (774.8)
Median (IQR)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)	0 (0–0)
Range	0–5’000	0–13’000	0–500	0–7’090	0–11’800	0–73’240	0–4’800
missing, no (%)	*1 (2%)*	*2 (4%)*	*1 (2%)*	-	-	*2 (5%)*	-
Employment and income	**Parents n=107**	**Parents n=99**	**Parents n=96**	**Parents n=92**	**Parents n=86**	**Parents n=78**	**Parents n=75**
Full-time equivalent unit							
Mean (SD)	0.6 (0.4)	0.6 (0.4)	0.6 (0.4)	0.6 (0.4)	0.6 (0.4)	0.6 (0.4)	0.6 (0.4)
Median (IQR)	0.6 (0.2–1.0)	0.8 (0.3–1.0)	0.8 (0.3–1.0)	0.7 (0.2–1.0)	0.8 (0.2–1.0)	0.6 (0.1–1.0)	0.7 (0.2–1.0)
Range	0–1.4	0–1.4	0–1.4	0–1.4	0–1.4	0–1.4	0–1.4
missing, n (%)	*1 (1%)*	*7 (7%)*	*7 (7%)*	*5 (5%)*	*4 (5%)*	*5 (6%)*	*4 (5%)*

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Outcome	Day 30	Day 60	Day 90	Day 120	Day 150	Day 240	Day 330
Income in CHF, n (%)							
No income	14 (13%)	12 (12%)	12 (13%)	13 (14%)	12 (14%)	12 (15%)	10 (13%)
1–2'999	19 (18%)	16 (16%)	15 (16%)	16 (17%)	16 (19%)	13 (17%)	12 (16%)
3'000–3'999	9 (8%)	7 (7%)	6 (6%)	6 (7%)	4 (5%)	7 (9%)	5 (7%)
4'000–4'999	12 (11%)	11 (11%)	10 (10%)	9 (10%)	10 (12%)	7 (9%)	9 (12%)
5'000–5'999	15 (14%)	14 (14%)	13 (14%)	13 (14%)	12 (14%)	11 (14%)	12 (16%)
6'000–6'999	14 (13%)	11 (11%)	11 (11%)	9 (10%)	9 (10%)	8 (10%)	8 (11%)
7'000–7'999	3 (3%)	3 (3%)	3 (3%)	4 (4%)	4 (5%)	4 (5%)	3 (4%)
8'000–8'999	4 (4%)	3 (3%)	3 (3%)	3 (3%)	3 (3%)	2 (3%)	2 (3%)
9'000–10'000	5 (5%)	4 (4%)	4 (4%)	4 (4%)	4 (5%)	3 (4%)	3 (4%)
>10'000	7 (7%)	7 (7%)	7 (7%)	6 (7%)	5 (6%)	5 (6%)	5 (7%)
missing	*5 (5%)*	*11 (11%)*	*12 (13%)*	*9 (10%)*	*7 (8%)*	*6 (8%)*	*6 (8%)*
Work absenteeism, in days	**Parents in employment[a] n=88**	**Parents in employment[a] n=77**	**Parents in employment[a] n=74**	**Parents in employment[a] n=69**	**Parents in employment[a] n=65**	**Parents in employment[a] n=55**	**Parents in employment[a] n=56**
Sick leave, n (%)							
0	49 (56%)	46 (60%)	51 (69%)	40 (58%)	35 (54%)	30 (55%)	28 (50%)
1–5	12 (14 %)	9 (12%)	6 (8%)	7 (10%)	9 (14%)	11 (20%)	16 (29%)
6–10	2 (2%)	3 (4%)	6 (8%)	4 (6%)	4 (6%)	1 (2%)	1 (2%)
11–15	3 (3 %)	2 (3%)	2 (3%)	2 (3%)	3 (5%)	2 (4%)	1 (2%)
>15	6 (7 %)	3 (4%)	2 (3%)	3 (4%)	3 (5%)	1 (2%)	1 (2%)
missing	*16 (18 %)*	*14 (18%)*	*7 (9%)*	*13 (19%)*	*11 (17%)*	*10 (18%)*	*9 (16%)*
Vacation, n (%)							
0	43 (49%)	38 (49%)	37 (50%)	37 (54%)	33 (51%)	13 (24%)	20 (36%)
1–5	18 (20%)	16 (21%)	10 (14%)	12 (17%)	17 (26%)	15 (27%)	12 (21%)
6–10	5 (6%)	4 (5%)	11 (15%)	7 (10%)	2 (3%)	10 (18%)	8 (14%)
11–15	2 (2%)	3 (4%)	2 (3%)	1 (1%)	-	2 (4%)	3 (5%)
>15	2 (2%)	-	2 (3%)	1 (1%)	-	5 (9%)	2 (4%)
missing	*18 (20%)*	*16 (21%)*	*12 (16%)*	*11 (16%)*	*13 (20%)*	*10 (18%)*	*11 (20%)*

SD indicates standard deviation; IQR, inter quartile range; CHF, Swiss francs.
[a] Number of parents for which full-time-equivalent units were available. Some parents started to work again, explaining the increase in n at day 330.

Supplemental Table 4: Results of Little's missing completely at random test.

Outcome variable group	*p*
Out-of-pocket expenses	*0.82*[a]
Employment and income	*0.005*[b]
Work absenteeism	*<0.001*[b]

[a] A p-value of >0.05 suggests that there is no significant evidence to reject the null hypothesis of missing completely at random. The missing data is likely to be missing completely at random.
[b] <0.05. The missing data is likely to be missing not at random.

Supplemental Table 5: Financial support received by participating families during the 330-day care follow-up assessment.

Financial support in CHF	**Day 30**	**Day 60**	**Day 90**	**Day 120**	**Day 150**	**Day 240**	**Day 330**
	Families n=59	Families n=54	Families n=52	Families n=50	Families n=47	Families n=42	Families n=40
Financial support, n (%)							
0	40 (68%)	31 (57%)	28 (54%)	31 (62%)	28 (60%)	21 (50%)	21 (53%)
1–499	4 (7%)	4 (7%)	5 (10%)	4 (8%)	5 (11%)	2 (5%)	1 (3%)
500–999	-	2 (4%)	3 (6%)	1 (2%)	2 (4%)	2 (5%)	1 (3%)
1'000–1'999	4 (7%)	3 (6%)	3 (6%)	4 (8%)	3 (6%)	1 (2%)	1 (3%)
2'000–2'999	5 (8%)	7 (13%)	4 (8%)	5 (10%)	5 (11%)	3 (7%)	3 (8%)
3'000–5'000	3 (5%)	1 (2%)	2 (4%)	-	1 (2%)	3 (7%)	3 (8%)
>5'000	1 (2%)	2 (4%)	5 (10%)	3 (6%)	1 (2%)	7 (17%)	10 (25%)
missing	*2 (3%)*	*4 (7%)*	*2 (4%)*	*2 (4%)*	*2 (4%)*	*3 (7%)*	-

CHF indicates Swiss francs.

Supplementary Table 6: Outcomes of the 300-day bereavement follow-up.

Outcome	**Day 120** Parents n = 23	**Day 330** Parents n = 23
Change in full-time-equivalent unit, n (%)		
No change	17 (74%)	20 (87%)
Increase	5 (17%)	1 (4%)
Decrease	1 (4%)	0 (%)
missing	-	*2 (9%)*
Change in income, n (%)		
No change	17 (74%)	20 (87%)
Increase	5 (17%)	0 (%)
Decrease	1 (4%)	1 (4%)
missing	*1 (4%)*	*2 (9%)*
Sick leave days, n (%)		
Not in employment	1 (4%)	1 (4%)
0	10 (44%)	12 (52%)
1–10	1 (4%)	1 (4%)
11–20	1 (4%)	1 (4%)
21–30	1 (4%)	1 (4%)
>30	4 (17%)	-
missing	*5 (22%)*	*7 (30%)*
Vacation days, n (%)		
Not in employment	1 (4%)	1 (4%)
0	9 (39%)	3 (13%)
1–10	5 (22%)	10 (39%)
11–20	1 (4%)	4 (17%)
21–30	-	-
>30	2 (9%)	-
missing	*5 (22%)*	*5 (22%)*

Supplemental Table 7: Complete case analyses of the effect of a child's hospital LoS on families' OOP expenses (missing completely at random) using adjusted two-way linear fixed-effects models.

Outcome	Complete-case adjusted models[a]			
	Observations	**Coef.**	**[95% CI]**	***p***
Out-of-pocket expenses				
Home healthcare supplies[b]	316	-1.47	[-3.57 to 0.63]	*0.17*
Travel and accommodation[b]	313	4.55	[2.50 to 6.60]	*<0.001*
Childcare and home help[b]	316	-3.05	[-12.00 to 5.89]	*0.50*
Special and extraordinary[b]	309	-64.09	[-182.80 to 54.01]	*0.29*

Coef. indicates coefficient; CI, confidence interval.
[a] The adjusted models are linear fixed-effects models with time and subject fixed effects adjusted for financial support and follow-up assessment period length.
[b] in Swiss francs.

Supplemental Table 8: Subgroup analysis by study participation using adjusted fixed-effects models.

Outcome	Participated for <330 days, i.e., drop-out, death of the child				Participated for the full 330 days			
	Observations[c]	Coef.	[95% CI]	*p*	Observations[c]	Coef.	[95% CI]	*p*
Out-of-pocket expenses								
Home healthcare supplies[a]	59	-9.19	[-26.39 to 8.00]	*0.30*	270	-1.04	[-2.99 to 0.91]	*0.30*
Travel and accommodation[a]	58	2.47	[-7.59 to 12.52]	*0.63*	270	4.04	[1.96 to 6.12]	*<0.001*
Childcare and home help[a]	59	-4.67	[-32.46 to 23.12]	*0.74*	270	-3.96	[-13.58 to 5.66]	*0.42*
Special and extraordinary[a]	59	-4.86	[-23.05 to 13.33]	*0.60*	269	-57.08	[-182.18 to 68.03]	*0.37*
Employment and income								
Full-time equivalent unit	96	0.00	[-0.01 to 0.01]	*0.52*	489	-0.00	[-0.00 to 0.00]	*0.98*
Income[a]	86	22.33	[-52.18 to 96.85]	*0.56*	478	2.53	[-7.43 to 12.50]	*0.62*
Work absenteeism								
Sick leave[b]	64	0.03	[-0.49 to 0.54]	*0.92*	337	0.03	[-0.02 to 0.09]	*0.19*
Vacation[b]	63	-0.21	[-0.53 to 0.11]	*0.22*	325	-0.06	[-0.14 to 0.02]	*0.12*

Coef. indicates coefficient; CI, confidence interval.
[a] in Swiss francs.
[b] in days.
[c] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).

Supplemental Table 9: Subgroup analysis by child hospital length of stay using adjusted fixed-effects models.

Outcome	Hospitalised for <15 days during study participation				Hospitalised for ≥15 days during study participation			
	Observations[c]	Coef.	[95% CI]	*p*	Observations[c]	Coef.	[95% CI]	*p*
Out-of-pocket expenses								
Home healthcare supplies[a]	191	-0.87	[-10.76 to 9.02]	*0.86*	138	-1.22	[-3.21 to 0.78]	*0.23*
Travel and accommodation[a]	190	12.62	[3.83 to 21.40]	*0.006*	138	3.21	[1.03 to 5.39]	*0.005*
Childcare and home help[a]	191	-5.92	[-48.46 to 36.62]	*0.79*	138	-5.12	[-14.00 to 3.75]	*0.26*
Special and extraordinary[a]	190	-438.68	[-1085.05 to 211.69]	*0.19*	138	6.74	[-14.36 to 27.83]	*0.53*
Employment and income								
Full-time equivalent unit	345	0.00	[-0.01 to 0.01]	*0.86*	240	0.00	[-0.00 to 0.00]	*0.76*
Income[a]	342	2.27	[-44.88 to 49.43]	*0.93*	222	1.18	[-6.69 to 9.04]	*0.77*
Work absenteeism								
Sick leave[b]	258	0.01	[-0.20 to 0.22]	*0.90*	143	0.04	[-0.03 to 0.11]	*0.23*
Vacation[b]	246	-0.14	[-0.47 to 0.19]	*0.40*	142	-0.05	[-0.13 to 0.04]	*0.27*

Coef. indicates coefficient; CI, confidence interval.
[a] in Swiss francs.
[b] in days.
[c] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).

Supplemental Table 10: Subgroup analysis by household income using adjusted fixed-effects models.

Outcome	Household income <CHF100'000				Household income ≥CHF100'000			
	Observations[c]	Coef.	[95% CI]	*p*	Observations[c]	Coef.	[95% CI]	*p*
Out-of-pocket expenses								
Home healthcare supplies[a]	145	-2.35	[-5.02 to 0.33]	*0.09*	176	-2.02	[-3.11 to 2.70]	*0.89*
Travel and accommodation[a]	145	6.67	[3.95 to 9.40]	*<0.001*	175	2.09	[-0.77 to 4.95]	*0.15*
Childcare and home help[a]	145	-0.97	[-2.81 to 0.88]	*0.31*	176	-6.51	[-21.46 to 8.44]	*0.40*
Special and extraordinary[a]	144	-75.39	[-232.08 to 81.31]	*0.35*	176	-38.52	[-207.79 to 130.75]	*0.66*
Employment and income								
Full-time equivalent unit	257	-0.00	[-0.00 to 0.00]	*0.12*	317	0.00	[-0.00 to 0.00]	*0.67*
Income[a]	258	6.49	[-1.16 to 14.14]	*0.10*	300	-2.00	[-19.57 to 15.56]	*0.82*
Work absenteeism								
Sick leave[b]	149	0.04	[-0.05 to 0.14]	*0.40*	250	0.03	[-0.04 to 0.09]	*0.41*
Vacation[b]	152	-0.07	[-0.19 to 0.04]	*0.18*	234	-0.05	[-0.14 to 0.05]	*0.32*

Coef. indicates coefficient; CI, confidence interval; CHF, Swiss francs.
[a] in Swiss francs.
[b] in days.
[c] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).

Supplemental Table 11: Subgroup analysis by diagnosis using adjusted fixed-effects models.

Outcome	Non-neurological				Neurological			
	Observations[b]	**Coef.**	**[95% CI]**	***p***	**Observations**[b]	**Coef.**	**[95% CI]**	***p***
Out-of-pocket expenses								
Home healthcare supplies[a]	89	-2.82	[-6.08 to 0.45]	*0.10*	240	-0.65	[-3.40 to 2.10]	*0.64*
Special and extraordinary[a]	88	-113.99	[-351.44 to 123.47]	*0.35*	240	-57.68	[-193.37 to 78.01]	*0.41*

Coef. indicates coefficient; CI, confidence interval.
[a] in Swiss francs.
[b] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).

Supplemental Table 12: Subgroup analysis by home-to-hospital travel distance using adjusted fixed-effects models.

Outcome	Home-hospital distance <21 kilometres				Home-hospital distance ≥21 kilometres			
	Observations[b]	Coef.	[95% CI]	*p*	Observations[b]	Coef.	[95% CI]	*p*
Out-of-pocket expenses								
Travel and accommodation[a]	120	1.66	[-0.89 to 4.20]	*0.21*	208	4.85	[2.14 to 7.55]	*<0.001*

Coef. indicates coefficient; CI, confidence interval.
[a] in Swiss francs.
[b] Number of observations vary because of variation in missing outcome and financial support data (see Supplemental Tables 3 and 5 for information on missing data).

Chapter 4: The Funding of Specialised Paediatric Palliative Care in Switzerland: A Conceptualisation and Modified Delphi Study on Obstacles and Priorities

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4.1 Highlights

- Funding limitations likely pose a major barrier to the provision of specialised paediatric palliative care (SPPC) in Switzerland. However, evidence on existing models of funding as well as their shortcomings and strategies for improvements are scarce. The available evidence suggests both that reimbursement mechanisms tend to under-value care input, and that funding models are often characterised by a combination of public, private and philanthropic funding.
- Our conceptualisation of the direct financial flows and funding arrangements regarding hospital-based consultative SPPC programmes in Switzerland visualises the complexity and fragmentation in the current models of funding. By conducting a three-round modified Delphi study, 23 obstacles to and 29 priorities for funding SPPC programmes sustainably were identified. The obstacles and priorities indicate that improvements in SPPC-related funding models are needed to ensure that the needs of affected children and their families are adequately met.

4.2 Abstract

Background

Effective funding models are key for implementing and sustaining critical care delivery programmes such as specialised paediatric palliative care (SPPC). In Switzerland, funding concerns have frequently been raised as primary barriers to providing SPPC. However, evidence on existing models of funding, their shortcomings and strategies for improvements remains scarce.

Aims

This study's first aim was to investigate and conceptualise the funding of hospital-based consultative SPPC programmes in Switzerland. Its second aim was to identify obstacles to and priorities for funding these programmes sustainably.

Methods

A four-step process, including a document analysis, was used to conceptualise the funding of hospital-based consultative SPPC programmes in Switzerland. In consultation with a

purposefully selected panel of experts in the subject, a three-round modified Delphi study was conducted to identify funding-relevant obstacles and priorities regarding SPPC.

Results

Current funding of hospital-based consultative SPPC programmes is complex and fragmented, combining funding from public, private and charitable sources. Overall, 21 experts participated in the first round of the modified Delphi study, 19 in round two and 15 in round three. They identified 23 obstacles and 29 priorities. Consensus (>70%) was obtained for 12 obstacles and 22 priorities. The highest level of consensus (>90%) was achieved for three priorities: the development of financing solutions to ensure long-term funding of SPPC programmes; the provision of funding and support for integrated palliative care; and sufficient reimbursement of inpatient service costs in the context of high-deficit palliative care patients.

Conclusions

Decision and policy makers hoping to develop and expand SPPC in Switzerland should be aware that current funding models are highly complex and that the current funding of SPPC is impeded by several obstacles. Considering the steadily rising prevalence of children with life-limiting conditions and the benefits of SPPC, improvements in funding models are needed to ensure that SPPC programmes fully meet the needs of affected children and their families.

4.3 Introduction

The prevalence of children (aged 0–19 years) with life-limiting conditions has been rising steadily in recent years.[1,2] Extrapolating from hospital admission data from England,[2] up to 11'200 children with LLCs are currently estimated to live in Switzerland. While advances in life-extending medical care and technology can partially explain the steady increase,[3-5] improvements in medical coding practice may also have had an effect.[2] Many of these children and their families can benefit from paediatric palliative care (PPC). As an active, holistic approach, PPC aims to improve the quality of life of children with severe health-related suffering, as well as their families and caregivers.[6] As a needs-based approach, PPC includes physical, emotional, social and spiritual elements that continue throughout the child's life and beyond.[6,7]

Recognising the complexity of care involved, the Swiss Federal Office of Public Health defines PPC as *specialised palliative care*.[8] Ideally, it is provided by specialised PPC (SPPC) teams consisting of physicians, nurses, therapists and other professionals working exclusively in

PPC.[9,10] In Switzerland, SPPC is commonly provided by consultative hospital-based teams/programmes.[11-13] Although new programmes have been implemented in recent years, nationwide access remains limited.[11,12]

SPPC offered within a consultative model of care contributes to primary care provision and incorporates elements of medical treatment, care coordination, psychosocial support and other consultative services.[13] Care and support is offered as and where necessary—both inside and outside of hospitals (mobile services such as home visits), through the phases of palliation, end of life and bereavement—to patients, their families, primary care teams and other healthcare professionals.[13] The level of mobile support offered varies between programmes, depending on service mandates and available resources.[14] Additionally, SPPC teams may engage in PPC-related education, training and research.[13]

Although federal healthcare laws and regulations apply[15,16] and most cantons have formally recognised the promotion and provision of palliative care (PC),[17] SPPC is currently much less established than adult PC.[11] In the context of the Swiss healthcare system's complexity, for which federal and cantonal bodies assume different tasks, ongoing resource shortages are likely to challenge SPPC's full provision. In the Swiss healthcare system, resources to pay for eligible services, including PC, are collected mostly through compulsory insurance premiums and taxes.[18] In addition, patients who use insured services are subject to cost-sharing in the form of deductibles and co-payments.[18] And while patients under the age of 18 are exempt from deductible payments, their families are still liable for co-payments.[19]

Activity-based funding is the dominant payment method to reimburse healthcare providers in Switzerland.[18] While inpatient services are reimbursed via Diagnosis Related Group (DRG) payments, outpatient medical services are reimbursed via the tarif médical (TARMED), a fee-for-service system.[18] Reimbursement of inpatient costs is subject to cost-sharing between cantons (at least 55%) and health insurers (at most 45%).[18] Under certain circumstances, for example, when a child has a birth defect, the Swiss disability insurance covers part of the related healthcare expenses.[20] It reimburses 80% of inpatient treatment costs, with the canton of residence bearing the remaining 20%.[21] Payments for medical devices and items, laboratory and diagnostic services and medications are specified in standard fee schedules (i.e., so-called *positive lists*).[18] Although reimbursement via standardised payment systems, for example, SwissDRG and TARMED, may work well in most healthcare settings, this is not always the case for hospital-based consultative SPPC programmes. Considering these programmes'

complexity, with care and support provided in various settings and across the phases of palliation, end of life and bereavement, adequate reimbursement of related costs may constitute a major challenge.

Information on models of SPPC funding and their practical implementation remains scarce. Even in adult PC, few studies describe such models.[22] The available evidence suggests both that reimbursement mechanisms tend to undervalue care input, and that funding models are often characterised by a combination of public, private and philanthropic funding.[22] However, it has been recognised that analyses of payment and financial strategies based on programme types and funding systems are highly important to this fields progress.[23] Therefore, this study's primary aim was to develop a conceptual model describing the funding of hospital-based consultative SPPC programmes in Switzerland. Its second aim was to identify obstacles to and priorities for funding these programmes sustainably.

4.4 Materials and methods

In this study, two separate methodological approaches were employed to address the study's objectives. First, to develop a conceptual model describing the funding of hospital-based consultative SPPC programmes in Switzerland, we followed a four-step conceptualisation process, including a document analysis. Second, to identify obstacles and priorities regarding SPPC funding, we conducted a three-round modified Delphi study.

4.4.1 Conceptualisation process

Conceptual models provide visual illustrations of causal linkages (often visualised as arrows) among sets of concepts (often visualised as boxes) believed to relate to particular target points.[24,25] To conceptualise the funding of hospital-based consultative SPPC programmes in Switzerland, we used a four-step process: (1) defining a target point, (2) choosing a conceptual basis, (3) conducting a literature search and (4) proposing a conceptual model.[24]

Target point

For this conceptualisation, we decided to set the focus on hospital-based consultative SPPC programmes, as they are a common model for providing PPC and have been implemented at several children's hospitals throughout Switzerland.[11,12]

Conceptual basis

Deber et al.'s[26] blended service and funding flow model provided the theoretical basis upon which we conceptualised the funding of hospital-based consultative SPPC programmes in Switzerland. Their model illustrates the complex relationship between provider organisations, service providers, service recipients and third-party payers, all of which are connected by payment and reimbursement structures.[26] To design our model, we focused on assessing the current sources of funding, systems of payment and mechanisms of reimbursement in terms of direct financial flows and funding arrangements.

Document analysis

To explore and describe the funding of Swiss SPPC programmes, we performed a document analysis. Such analyses are widely used in health policy research to review documents, provide context and supplement other data types.[27] The aim of our document analysis was to identify funding sources, payment systems and reimbursement mechanisms, and to uncover areas where challenges to SPPC programmes' funding are encountered.

Due to the limited number of SPPC-specific documents and the fact that healthcare financing policies do not distinguish between PC for children and adults, we widened our search to include documents on the funding of PC in general. The READ approach (Ready materials; Extract data; Analyse data; and Distil findings) for document analysis in health policy research provided the necessary methodology for this analysis.[28]

Documents were identified by conducting web-searches for grey literature (Google search engine), browsing for documents on institutions' and non-governmental organisations' web-pages and tracking references. The search was conducted in German between August 6 and 20, 2021 by the first author (S.M.) and discussed with two other authors (K.Z. and E.B). Documents about PC funding in Switzerland were included if they reported funding sources, payment systems and reimbursement mechanisms and/or areas where challenges to that funding are encountered. To account for the implementation of SwissDRG, documents had to be published in 2012 or later. Documents reporting solely on the funding of non-hospital-based PC (e.g., geriatric long-term care, home care agencies) were excluded. Included documents were analysed using qualitative content analysis.[27] Information was coded, summarised and tabulated into three predefined categories: funding sources; payment systems and reimbursement mechanisms; and areas of challenges.

Conceptual model

The document analysis' findings were used to visualise the funding of hospital-based consultative SPPC programmes. The resulting conceptual model is presented in Figure 1.

4.4.2 Modified Delphi study

To address our second aim, that is, to identify obstacles and priorities in the funding of Swiss SPPC programmes, we used a three-round modified Delphi approach.[29] The Delphi technique is a well-established, iterative series of steps to survey experts on a particular issue and develop individual opinions into group consensus.[30-34] Since its inception in the 1950s, numerous versions of the Delphi technique have been developed, differing mainly regarding how consensus was reached or measured.[35] In this study, consensus was measured by asking participating experts to indicate their agreement or disagreement with specific statements on a four-point Likert scale.

Procedures

Before study start, we purposefully compiled an initial list of experts. We selected potential participants based on two inclusion criteria: either they had professional experience in establishing, managing or leading Swiss SPPC programmes (e.g., programme directors), or they had direct professional knowledge about sources of funding, payment systems or reimbursement mechanisms related to PC funding in Switzerland (e.g., researchers, health economists, public health officials, insurance professionals). All eligible experts were first invited via e-mail to participate in this study, then asked to name two to three other experts who would might also qualify for participation. The modified Delphi study took place between 23 May and 3 October, 2022. Online questionnaires were provided in German via Google Forms. Reminders were sent towards the end of each round.

Round one

All experts who had consented to study participation received a link to an online questionnaire. This asked them to provide minimal demographics (i.e., profession, affiliation) and, using free-text fields, to list and describe obstacles and priorities regarding SPPC funding. To provide the necessary context for their responses, areas of challenges identified via the document analysis were provided. In discussion with two study team members (K.Z. and E.B.), the first author

(S.M.) compiled, summarised and merged the experts' answers into two lists, one of obstacles, the other of priorities.

Round two

In round two, the generated and anonymised lists were sent to everyone who had participated in round one. Experts were encouraged to comment on identified obstacles and priorities, as well as to specify any more that might have emerged or occurred to them during the second round. Their answers were again compiled, summarised and merged. The results were used to update the lists of obstacles and priorities.

Round three

To measure consensus, the updated lists were sent to all experts who had participated in the first two rounds. In the third questionnaire, experts were asked to indicate their agreement or disagreement with each identified obstacle and priority on a four-point Likert scale: strongly agree, agree, disagree, strongly disagree. In cases where participating experts did not feel adequately informed on a topic to indicate agreement or disagreement, they were given the option to answer *don't know*. In addition, each respondent was asked to indicate what they considered the three most pressing obstacles and the three most urgent priorities.

Data analysis

Descriptive statistics (i.e., count and percent) were used to provide an overview of the characteristics of participating experts and to examine consensus. The analyses were performed in Microsoft Excel. Thresholds of consensus vary widely between Delphi studies, with 75% being the median.[36] Considering the diversity of our contributors, we chose a slightly lower cut-off, defining consensus as >70% agreement (strongly agree and agree).

4.5 Results

4.5.1 Document analysis and conceptual model

We included a total of 15 documents in our analysis: 10 reports,[11,12,14,16,17,37-41] three technical articles,[42-44] one directive[45] and one review.[46] Twelve were obtained through web searches and three by backward reference tracking. Only two were paediatric-specific; the other 13 focused on PC in general. An overview of included documents is provided as a Supplemental Material.

Figure 1 shows our conceptual model of hospital-based consultative SPPC programme funding in Switzerland.

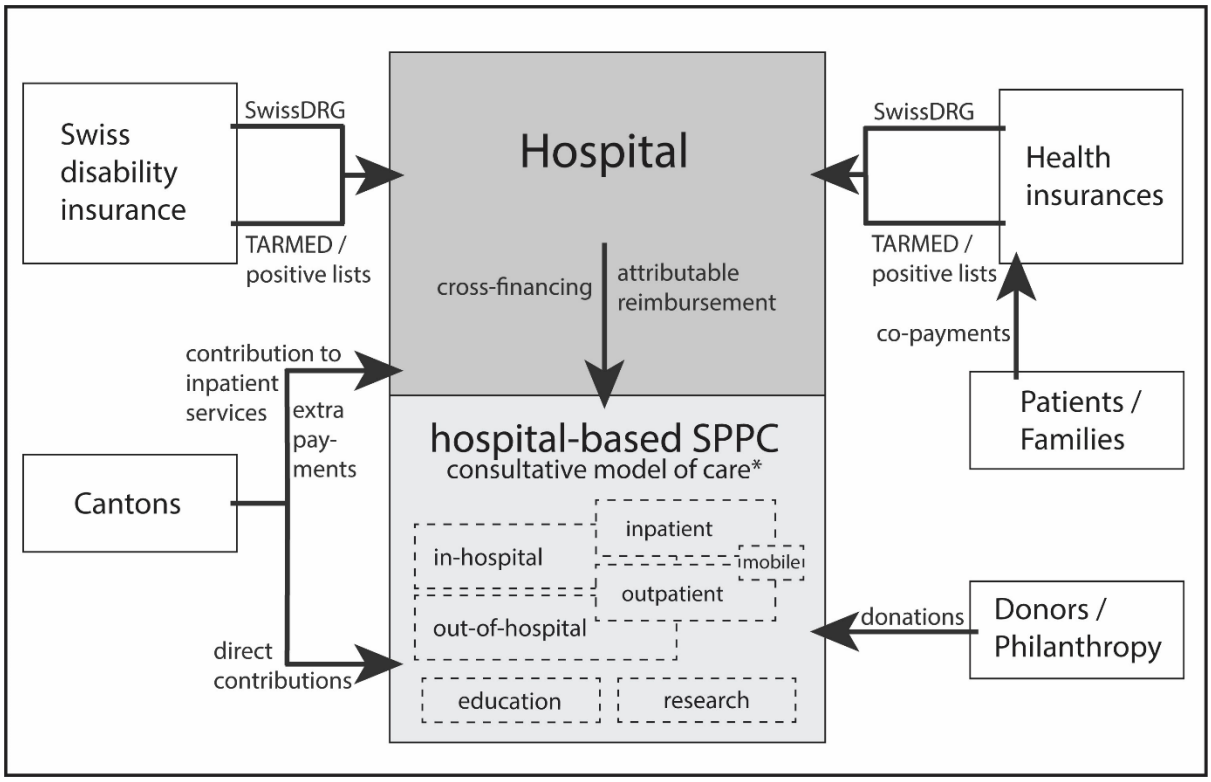

Figure 1: Conceptualisation of direct financial flows and funding arrangements regarding hospital-based consultative SPPC programmes in Switzerland. *The consultative model of care refers to the provision of medical treatment, care coordination, psychosocial support and other consultative services that contribute to primary care provision. Care and support is provided to families, primary care teams and other professionals in- and outside of hospitals, i.e., inpatient, outpatient/mobile services. SPPC teams may engage in PPC-related research and education.

Funding sources

Figure 1 illustrates how the Swiss disability insurance, health insurances, cantons, donors and philanthropists, patients (i.e., their families) and hospitals all hold stakes in the funding of hospital-based consultative SPPC programmes.[11,12,14,16,17,41] Depending on patient characteristics (e.g., age, diagnosis) and service type (e.g., medical aid, treatment), certain costs are reimbursed (partially) either by the Swiss disability insurance or by health insurers.[14,16]

In addition to partially reimbursing inpatient service costs and providing financial grants to service providers (e.g., extra payments, deficit coverage), some cantons provide direct financial contributions to fund PC.[11,12,14,16,17] In these cantons, special service mandates with hospitals

regulate PC provision and related cantonal funding.[17] For instance, the canton of Vaud commissioned a cantonally-funded consultative mobile SPPC programme to provide care in various settings, for example, hospitals, long-term care institutions and patients' homes.[11,12,16]

Very limited information is reported about payments made by patients or families (in case of children). Leaving aside their tax payments and insurance premiums, however, they are also involved in the financing of PC services through their co-payments.[14]

For hospital-based programmes, hospitals act as both funders and distributors of funds generated through service provision.[41] I.e., payments made by the Swiss disability insurance, health insurances and cantons are collected at the hospital level and distributed under the sovereignty of the hospital.[41] Our document analysis indicated that SPPC programmes' budget deficits had to be either cross-financed by their operating hospitals[37,46] or covered by donations and philanthropic contributions.[11,17,37,46]

Payment systems and reimbursement mechanisms

In terms of the payment systems and reimbursement mechanisms outlined in Figure 1, the reviewed documents primarily reported on SwissDRG and TARMED. The responsibility for further developing, adjusting and maintaining the SwissDRG system belongs to the SwissDRG AG, a non-profit public organisation and joint institution of healthcare provider associations, health insurers and cantons.[42] Within the National Strategy for Palliative Care 2010 – 2015[40], SwissDRG AG was commissioned to develop a national tariff structure for reimbursing inpatient PC services.[42] Via a multi-year process, they developed Swiss Classification of Operations (CHOP) codes for PC procedures.[16,41-43,46] To ensure uniform, high-quality service provision, minimum structural and personnel requirements were set as performance criteria.[16,41-43] Only when these criteria are met can a hospital code and bill for the associated PC procedure codes.[41,42]

Despite these provisions, analyses showed that certain characteristics of PC patients were not being considered optimally regarding reimbursement.[42] For instance, in terms of lengths of stay, treatment costs and number of hospitalisations, PC patients differed significantly from other patients in the same DRGs.[42] Therefore, SwissDRG AG conducted a fundamental 'grouper' restructuring and classified PC as a pre-Major Diagnostic Category.[12,16,42] By the end of 2016, PC had been allocated a separate diagnosis group—one independent from the patient's main diagnosis, with its own DRG codes (codes A97A-G)—defined in terms of medical treatment, procedure, length of stay and other criteria.[42-44] Concerns about adverse incentives

regarding inappropriately shortened hospital stays were addressed by a two-pronged strategy: on one side, additional payments were allowed for extended hospitalisations;[45] on the other, case-consolidations were prevented by recognising readmissions as new, separate hospitalisations.[16,43]

Outpatient medical PC services are reimbursed on a fee-for-service basis via TARMED.[14,41] Services listed in this tariff structure are covered by compulsory healthcare insurance. However, our document analysis suggests that certain PC services, for example, care coordination and case management, are not fully reimbursable via TARMED.[16,41] Alongside TARMED, standard fee schedules—*positive lists*—regulate and ensure the reimbursement of diagnostic and laboratory services, medical devices and items, medications and other expenses (e.g., therapies).[16] Our document analysis also drew our attention to essential PC services, including support and relief for relatives and social counselling, that were not covered by formal payers.[14,16,41] Instead, these services must be financed by public service contracts, grants, donations or cross-financing.[14]

Areas of challenge

The reviewed documents indicate that funding challenges are a major barrier to the implementation, sustainability and further development of SPPC programmes.[11,40] Difficulties attached to charging for services/covering costs lead to deficits and funding gaps.[11,12,17,46] Areas where funding challenges are encountered include both the SwissDRG and the TARMED system.[12,14,16,37,38,40,41,43,44] In addition, reviewed documents suggest that variations between cantonal funding regulations hinder the provision of inter-cantonal mobile PC services.[17,37,40] Moreover, dependency on external funding (e.g., donations, philanthropic contributions) poses a risk to long-term financial stability.[17,41]

4.5.2 Modified Delphi study

Thirty-one experts were invited for study participation, of whom 22 had been purposefully identified by the study team and nine recommended by initially-contacted experts. Overall, 21 participated in the first Delphi round (68% response rate), 19 in the second (10% dropout rate) and 15 in the third (21% dropout rate). Demographic characteristics of the original 21 participating experts are presented in Table 1. The majority of participants were female (n=11, 52%), aged 50–69 years (n=11, 52%) and working in (university) hospitals, clinics or other healthcare

providers (n=14, 67%). Ten (48%) worked in medical/clinical professions, including SPPC programme leadership.

At the end of round one, one list each of obstacles (n=22) and priorities (n=28) regarding the funding of SPPC programmes was generated. After additional obstacles and priorities suggested in round two, round three began with lists of 23 obstacles and 29 priorities. Obstacles and priorities were grouped inductively into six categories: (1) *political and structural*, (2) *funding and tariff structures in general*, (3) *inpatient tariff structures*, (4) *outpatient tariff structures*, (5) *mobile PC* and (6) *other*. All identified obstacles and priorities are presented in Tables 2 and 3 respectively.

Table 1: Demographic characteristics of participating experts.

Characteristics	Experts, n=21
Gender, n (%)	
Female	11 (52%)
Male	10 (48%)
Age, n (%)	
30-49	10 (48%)
50-69	11 (52%)
Primary profession, n (%)	
Medical, clinical, SPPC programme leadership	10 (48%)
Health policy, health economics, public health	5 (24%)
Specialist in medical coding, payment systems, service reimbursement	4 (19%)
Research	2 (10%)
Primary place of employment, n (%)	
(University-)hospital, clinic, healthcare provider	14 (67%)
Federal office, (semi-)governmental organisation	3 (14%)
Association (e.g., hospital/insurance association)	2 (10%)
Other	2 (10%)

Obstacles

Using our predefined consensus definition of >70% of experts either strongly agreeing or agreeing (level of agreement), consensus was obtained on 12 of the 23 identified obstacles. A level of agreement of >85% was obtained for four obstacles; the absence of a holistic health policy approach; cantonal differences in service mandates and cost-coverage; a lack of PC-specific reimbursement codes in outpatient tariff structures; and existing consultation time limitations in the reimbursement of certain PC outpatient services in TARMED. Zero disagreement was recorded regarding difficulties arising from cantonal differences in PC service mandates

and cost-coverage. The distribution of identified obstacles' levels of agreement is shown in Figure 2.

Asked to indicate the three most pressing obstacles, participating experts indicated 18 obstacles at least one time to be most pressing. The obstacle most frequently named—six times—was the absence of a holistic health policy approach. The fragmentation of PC funding and a lack of guaranteed funding for developing and implementing new SPPC programmes were indicated four times each. Obstacles indicated as one of the most pressing fell predominantly within the political and structural category (Table 2).

Table 2: List of obstacles encountered in the funding of SPPC, sorted by category and most pressing.

Category	Obstacles encountered in the funding of SPPC	Most pressing
Political and structural	1. No holistic health policy approach to the financing and funding of PC[a]	6x
	2. Fragmentation of PC[a] funding rendering the establishment and maintenance of integrated and well-performing treatment chains more difficult	4x
	3. Lack of guaranteed funding to develop and implement SPPC programmes	4x
	4. Lack of legal definition of PC[a] (e.g., services, providers and funding needed to meet patients' PC[a] demand)	3x
	5. Dependency on charitable funding, compromising long-term continuity and sustainability of SPPC programmes	2x
	6. Cantonal differences regarding PC[a] service mandates and cost coverage (e.g., financial contributions, coverage of residual costs)	1x
Funding and tariff structures in general	7. Gaps in PC[a] funding when patients transition between care settings (e.g., inpatient, outpatient, home, rehabilitation)	3x
	8. Insufficient compensation, billing limitations and lack of tariffs regarding certain PC[a] services (e.g., roundtable meetings, case management, care coordination, support for relatives)	2x
	9. Patient classification and reimbursement difficulties arising from existing tariff structures that fail to recognise the heterogeneity, multimorbidity and complexity of the PC[a] population	-
Inpatient tariff structures	10. Insufficient reimbursement of inpatient service costs in the context of high-deficit PC[a] cases or PC[a] patients with complex case constellations (i.e., high-deficit outliers)	1x
	11. Difficulties in meeting the minimum criteria required for the PC Complex Codes of the Swiss Classification of Operations (CHOP)	1x
	12. Funding challenges due to gaps between remuneration and hospital operating costs in view of above-average operating costs, non-optimised processes, low base rates or gaps in tariff structures	-
Outpatient tariff structures	13. Lack of PC[a]-specific reimbursement codes in outpatient tariff structures	2x
	14. No reimbursement of bereavement support services or follow-up home visits to bereaved families and caregivers	2x
	15. Time limitations in the reimbursement of outpatient PC[a] services (i.e., consultation time limits in TARMED for cases not reimbursed via the Swiss disability, accident or military social insurances)	-
	16. Lack of clarity whether TARDOC, as a potential successor of TARMED, will improve PC[a] service reimbursement (TARDOC contains tariff positions for PC services provided by general practitioners and paediatricians)	-
Mobile PC	17. Difficulties in funding mobile PC[a] services, particularly non-direct patient services (e.g., care coordination, consultations with other healthcare professionals)	3x

continued on next page

Category	Obstacles encountered in the funding of SPPC	Most pressing
Other	18. Lack of financial support and relief for families and informal caregivers	3x
	19. Challenges to PC[a] service reimbursement in long-term and home-care settings	2x
	20. Inconsistent definitions of PC cases and populations (e.g., neonates, children, adolescents, adults, elderly) in discussions of funding issues	1x
	21. Lack of educational and training opportunities in SPPC	1x
	22. Insufficient evidence on SPPC's (cost-)effectiveness in the Swiss setting	1x
	23. Lack of national regulations about the inclusion, status and funding of (paediatric) hospices	-

PC indicates palliative care; SPPC, specialised paediatric palliative care; CHOP, Schweizerische Operationsklassifikation.

[a] Including but not limited to SPPC.

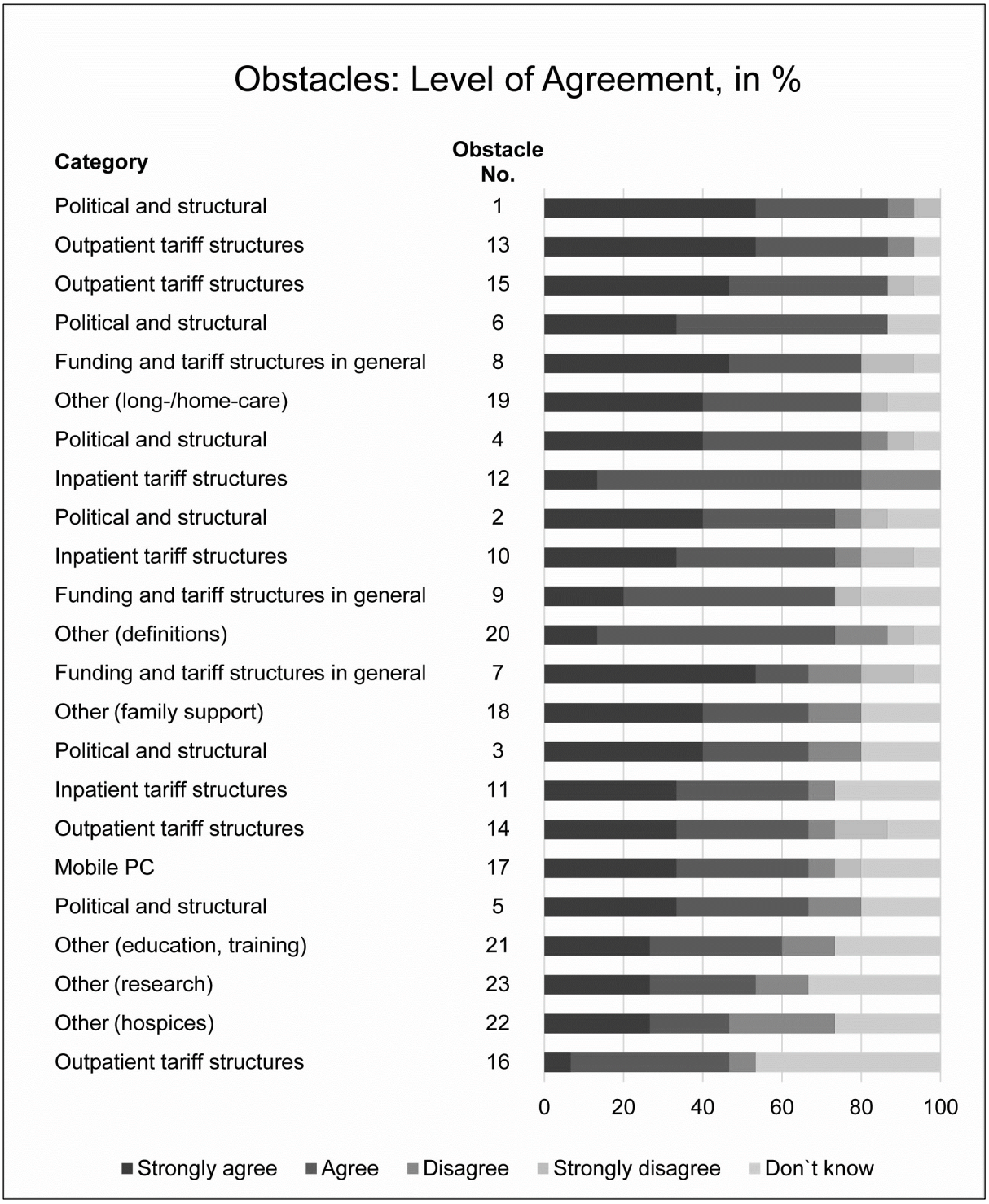

Figure 2: Distribution of the level of agreement on identified obstacles encountered in SPPC funding in Switzerland, sorted by level of agreement. Obstacle numbers refer to the numbered obstacles provided in Table 2.

Priorities

A level of agreement of >70% was measured for 22 of the 29 identified priorities. Three priorities had consensus rates of >90%: the development of financing solutions to ensure the long-term funding of SPPC programmes; the provision of funding and support for integrated PC programmes; and sufficient reimbursement of inpatient service costs in the context of high-deficit PC cases. Zero disagreement was recorded for two priorities: establishing a valid nationwide data-base on PC provision; and offering funding and support for integrated PC programmes, well-performing treatment chains and closer cooperation and coordination among service providers. The distributions of agreement levels of identified priorities are provided in Figure 3.

When asked to indicate what they considered the three most urgent priorities, participating experts noted a wide range of priorities (n=20). Most experts' top priorities fit within the political and structural category: inter-cantonal harmonisation of PC regulations was indicated five times; and the development of financing solution to ensure long-term funding of SPPC services, legislative integration of PC and the financing and support of integrated PC programmes were indicated four times each. Additionally, experts indicated four times that they considered the establishment of a valid nationwide data-base on PC provision one of the most urgent priorities (Table 3).

Table 3: List of priorities in the funding of SPPC, sorted by category and most urgent.

Category	Priorities in the funding of SPPC	Most urgent
Political and structural	1. Harmonisation of PC[a] regulations (e.g., service mandates, financing) and closer inter-cantonal cooperation and coordination in PC[a] provision	5x
	2. Funding and support for integrated PC[a] programmes, well-performing treatment chains and closer cooperation and coordination among service providers	4x
	3. Development of specific, feasible and viable funding solutions to ensure long-term funding of SPPC programmes	4x
	4. Legislative integration of PC[a] into the Swiss Federal Health Insurance Act (KVG) and the Swiss Health Care Benefits Ordinance (KLV)	4x
	5. Initiation of a nationwide working group (incl. decision-making bodies) for securing long-term funding in SPPC	3x
	6. Provision of financial resources (initial funding, core funding) to establish SPPC programmes and facilitate nationwide coverage	2x
	7. Comprehensive analysis of PC[a] demand, supply and funding, including the identification and disclosure of potential gaps	2x
	8. Establishment of a legal framework for the reimbursement of consultative PC[a] services	1x
Funding and tariff structures in general	9. Amendment of PC[a] services (incl. psychosocial, spiritual services) as standard benefits in the Swiss Statutory Health Insurance (OKP) scheme	1x
	10. Clarification of open questions regarding the reimbursement of PC[a] services provided when patients transition between care settings (e.g., inpatient, outpatient, home, rehabilitation)	1x
	11. Revision, further development and supplementation of services provided in the patient's absence in established tariff structures (e.g., interprofessional meetings, case management, care coordination)	-
	12. A comprehensive, PC-specific revision of payment systems and reimbursement mechanisms to improve PC[a] funding conditions in the medium-term	-
	13. A flexible application of tariff rules, explicitly approved by formal payers, to improve PC[a] funding conditions in the short term (e.g., via analogous positions)	-
Inpatient tariff structures	14. Sufficient reimbursement of inpatient service costs in the context of high deficit PC[a] cases or PC[a] patients with complex case constellations (i.e., high deficit outliers)	1x
	15. Assuring the quality of data supplied by service providers to SwissDRG AG (when adequate data becomes available, PC[a] patient and service classification improvements in inpatient tariff structures can be realised through system maintenance)	-
	16. Broader application of PC DRG complex codes through the respective certification (quality label) of SPPC programmes	-
	17. Consideration of structural factors not currently considered regarding SwissDRG at the hospital and patient level in setting base rates	-

continued on next page

Category	Priorities in the funding of SPPC	Most urgent
Outpatient tariff structures	18. Sufficient, cost-covering reimbursement of outpatient PC[a] services (e.g., interprofessional meetings, travel time for home visits)	2x
	19. Reduction of quantity and time limitations in the reimbursement of outpatient PC[a] services (e.g., reduction of consultation time limits in TARMED)	2x
	20. Introduction of PC[a]-specific counselling and coordination fees in outpatient tariff structures (i.e., tariff codes for PC[a] case management)	1x
	21. Reimbursement for bereavement support services provided to families and caregivers	1x
Mobile PC	22. Establishment of mobile PC[a] funding regulations in all cantons	-
	23. Funding of mobile PC[a] services based on the area's PC[a] demand, not contingent on fluctuating case numbers	-
Other	24. Establishment of a valid nationwide data-base on PC[a] provision, in coordination with the Spitalstationäre Gesundheitsversorgung (SpiGes) project of the Federal Office of Public Health and the Federal Statistical Office	4x
	25. Financial support/relief for PC[a] patients' families and informal caregivers	3x
	26. Development of educational and training opportunities in the field of PC[a] (incl. medical curricula)	2x
	27. Facilitation of research on SPPC's (cost-)effectiveness	1x
	28. Furthering knowledge and understanding of tariff structures to optimise the coding and billing of PC[a] services	1x
	29. Ensuring cost-covering financing of PC[a] services in non-hospital settings (e.g., hospices, psychiatric clinics, long-term institutions)	-

PC indicates palliative care; SPPC, specialised paediatric palliative care; KVG, Bundesgesetz über die Krankenversicherung; KLV, Krankenpflege-Leistungsverordnung; OKP, Obligatorische Krankenpflegeversicherung; DRG, Diagnosis Related Groups
[a] Including but not limited to SPPC.

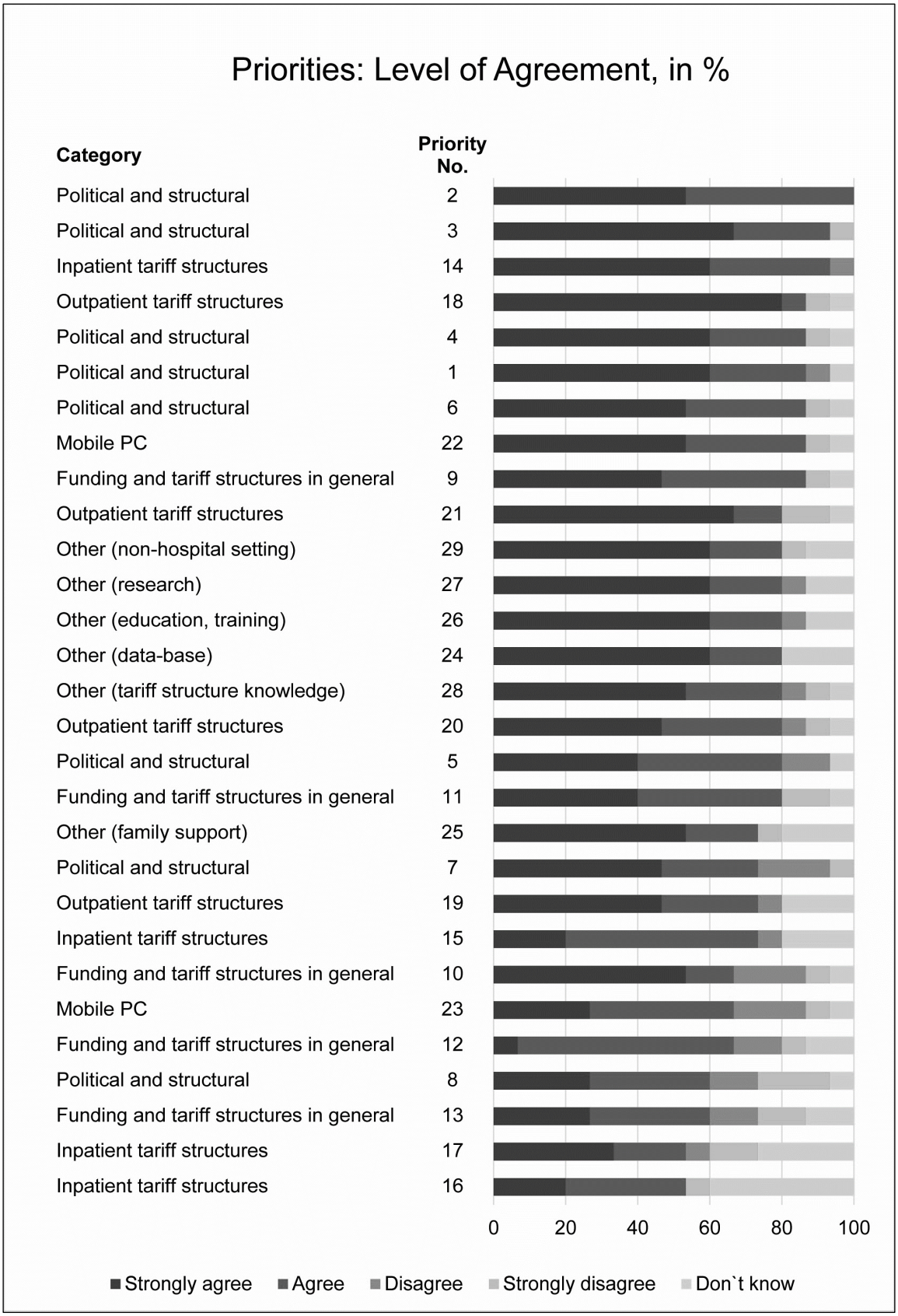

Figure 3: Distribution of the level of agreement on identified priorities for SPPC funding in Switzerland, sorted by level of agreement. Priority numbers refer to the numbered priorities provided in Table 3.

4.6 Discussion

Our conceptualisation of the funding of hospital-based consultative SPPC programmes in Switzerland (Figure 1) shows that funding flows and financial arrangements surrounding the provision of SPPC are highly fragmented. Our results indicate that donations and philanthropic contributions are required to supplement funding from formal structures. In addition, our modified Delphi study identified 23 obstacles and 29 priorities regarding SPPC funding. Consensus was reached on 12 of the obstacles and 22 of the priorities. The large numbers of both identified obstacles and priorities are notable, as both lists are samples of issues encountered in the funding of hospital-based consultative SPPC programmes. The contributing experts considered many of both lists as very pressing/urgent, suggesting that, while not one specific obstacle or priority emerged as most important, a bundle of each require urgent action.

This study's findings also bolster the results of a previous investigation on models of funding in PC.[22] Groeneveld et al.[22] suggest that a high degree of fragmentation in funding sources increases administrative complexity and create ambiguity in responsibilities. Fragmentations in funding sources, payment systems, and reimbursement mechanisms likely have serious implications for developing, implementing and sustaining SPPC programmes.

First, the findings of both our document analysis and our modified Delphi study suggest that high levels of administrative complexity in funding models hinder the development and implementation of new SPPC programmes. This would at least partially explain why nationwide coverage of SPPC has not yet been achieved in Switzerland.[11,12] Conversely, previous research has suggested that stable funding, with a straightforward system of payment and reimbursement greatly facilitates new programmes' establishment.[47]

Second, reliance on annual grants to cover operating costs can actually endanger a programme's long-term survival. For example, as both charitable foundations' and cantonal governments' budgets can freeze or dry up, dependence on either can pose serious risks to the long-term sustainability of SPPC programmes.

Third, fragmentation and complexity in SPPC programmes' funding may render it more difficult to estimate how and where funding sources, payment systems and reimbursement mechanisms act as policy levers. PC funding has recently become the focus of growing political attention in Switzerland.[41] In June 2021, parliamentary motion no. 20.4264[48] on PC financing

was passed, instructing the Federal Council to establish a statutory basis to guarantee needs-based palliative and end-of-life treatment and care for all people.[48]

Politically and structurally, the findings of our modified Delphi study suggest that further legislative integration and specification regarding PC funding is needed. In the case of motion no. 20.4264,[48] the parliament motion provides a unique opportunity to clarify open legal questions. Moreover, participating experts agreed that a nationwide working group should initiate work on securing long-term funding for SPPC. When executing parliamentary motion no. 20.4264,[48] the Federal Office of Public Health established two dedicated working groups: one for PC supply and demand, and one for PC financing. Ideally, these groups will commission a comprehensive analysis of SPPC demand, supply and funding. In addition to identifying potential gaps in SPPC supply, such an analysis would facilitate development of specific, viable long-term funding solutions.

Additionally, experts participating in our modified Delphi study agreed that differences in service mandates and funding regulations among Swiss cantons are an obstacle. Although most cantons have established legislation to promote PC,[17] the details of these measures are rather heterogenous. Therefore, even though tailored cantonal-level solutions for SPPC funding may provide flexibility in establishing new programmes, local differences may hamper the provision of inter-cantonal SPPC services (e.g., of mobile teams).

Our findings also indicate that charitable sources contribute disproportionately to the current funding of hospital-based consultative SPPC programmes in Switzerland: donations and philanthropic contributions cover up to 50% of annual budgets.[11] In this context, participating experts warned that reliance on donations and philanthropic contributions compromises SPPC programmes' long-term continuity and sustainability.

Regarding inpatient tariff structures, participating experts agreed that, in the context of high-deficit PC cases, that is, high-deficit outliers, improvements in the reimbursement of inpatient stay costs are required. Generally, compared with the total number of hospitalised patients, high-deficit outliers are a small number of patients that cause a substantial proportion of total inpatient stay costs.[49-51] Considering that SPPC cases are often highly complex,[52] high-deficit outliers can be expected to be more prevalent in this patient population. Sufficient reimbursement of these patients' treatment costs should thus be ensured.

Several obstacles and priorities identified in this study further indicate that certain PC activities are reimbursed insufficiently. These include but are not limited to care coordination, case management, consultations of other healthcare professionals and psychosocial and spiritual support. Previous research suggests that insufficient financing mechanisms constrain access to SPPC.[53,54] This issue is particularly evident in outpatient PC. One identified obstacle is that certain PC services are only partially billable, if at all, via outpatient tariff structures. Besides the services outlined above, those provided to family members and other informal caregivers, including psychosocial and bereavement support, are especially prone to reimbursement failures. Related issues regarding these services have also been documented previously.[14,16,38,41] Whether TARDOC, as a potential successor of TARMED, will improve the reimbursement of outpatient services provided by hospital-based consultative SPPC programmes remains unclear.

Strengths and limitations

The developed conceptual model provides a systemic understanding of how hospital-based consultative SPPC programmes are funded. While informing clinical and administrative leaders regarding the development and implementation of new SPPC programmes, it serves as a point of reference regarding funding issues, for example, how to address them through policies and regulations. Given that, with high levels of agreement among experts, we have identified a broad spectrum of obstacles and believe that our findings accurately reflect the issues encountered in SPPC funding. Initiatives aiming to improve SPPC funding models should focus on addressing the priorities identified above.

Several notable limitations affect this study. First, it was not always possible to strictly distinguish between SPPC and overall PC information. Therefore, we included documents on the funding of both SPPC and PC in general in our document analysis. Second, several experts participating in the modified Delphi study were experts not in SPPC but in PC funding. Third, our approach to conducting the modified Delphi study precluded us from defining the identified obstacles and priorities in greater detail. As a result, a number of obstacles and priorities are stated rather broadly. In addition, as this study aimed to quantify neither funding flows nor the financial impacts of identified obstacles and priorities, we recommend both topics as the subjects of further research. Future research may also explore funding issues in other settings, such as home-care and children's hospices.

4.7 Conclusions

Current funding of hospital-based consultative SPPC programmes in Switzerland is highly fragmented and characterised by complex combinations of public, private and charitable funding. With new SPPC programmes currently being developed and implemented, a systematic understanding of funding structures and requirements is critical. We are confident that our conceptual model as well as this study's identified obstacles and priorities will help researchers and policy makers understand the challenges the current system poses regarding SPPC funding. A clear view of these challenges can help them develop funding and reimbursement schemes that will appropriately support SPPC provision.

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4.9 Supplemental materials

Supplemental Table 1: Overview of included studies.

Type (year)	Publisher; author(s)	Title	Content category		
			Funding sources	Payment systems and reimbursement mechanisms	Areas of challenges
Report (2020)	Swiss Health Observatory; Peter C., Diebold M., Delgrande Jordan M., Dratva J., Kickbusch I., Stronski S.	Gesundheit in der Schweiz - Kinder, Jugendliche und junge Erwachsene: Nationaler Gesundheitsbericht 2020	X		X
Report (2020)	Amstad H.	Palliative Care für vulnerable Patientengruppen: Konzept zuhanden der Plattform Palliative Care des Bundesamtes für Gesundheit	X	X	X
Report / Strategy Paper (2012)	Bundesamt für Gesundheit BAG, Schweizerische Konferenz der kantonalen Gesundheitsdirektorinnen und -direktoren GDK	Nationale Strategie Palliative Care 2013–2015: Bilanz «Nationale Strategie Palliative Care 2010–2012» und Handlungsbedarf 2013–2015			X
Report (2020)	Bundesamt für Gesundheit BAG	Bessere Betreuung und Behandlung von Menschen am Lebensende: Bericht des Bundesrates in Erfüllung des Postulates 18.3384 der Kommission für soziale Sicherheit und Gesundheit des Ständerats (SGK-SR)	X	X	X
Report (2019)	Bundesamts für Gesundheit; Liechti L., Künzi K., Büro für arbeits- und sozialpolitische Studien BASS	Stand und Umsetzung von Palliative Care in den Kantonen: Ergebnisse der Befragung der Kantone und Sektionen von palliative ch 2018	X		X
Report (2013)	Bundesamt für Gesundheit BAG, Schweizerische Konferenz der kantonalen Gesundheitsdirektorinnen und -direktoren GDK; Furrer M.T., Grünig A., Coppex P.	Finanzierung der Palliative-Care-Leistungen der Grundversorgung und der spezialisierten Palliative Care (ambulante Pflege und Langzeitpflege)	X	X	X

continued on next page

Type (year)	Publisher; author(s)	Title	Content category		
			Funding sources	Payment systems and reimbursement mechanisms	Areas of challenges
Review (2018)	Gudat H.	Der Wert des Lebensendes: am Beispiel der Finanzierung der stationären spezialisierten Palliative Care in der Schweiz	X	X	X
Directive (2016)	SwissDRG AG	Beschluss des Verwaltungsrats der SwissDRG AG: Abbildung der palliativmedizinischen Behandlung im SwissDRG Tarifsystem		X	
Report (2020)	Degen E., Liebig B., Reeves E., Schweighoffer R.	Palliative Care in der Schweiz: Die Perspektive der Leistungserbringenden	X	X	X
Article (2016)	palliative ch; Schlägel F.	Das SwissDRG-System und die Finanzierung der palliativmedizinischen Versorgung		X	
Article (2016)	palliative ch; Gudat H.	Ist das Vergütungssystem der SwissDRG AG für spezialisierte Palliative Care geeignet? Die Pro-Position		X	X
Article (2016)	palliative ch; Borasio G.D.	Ist das Vergütungssystem der SwissDRG AG für spezialisierte Palliative Care geeignet? Die Kontra-Position		X	X
Report (2014)	Wächter M., Bommer A.	Mobile Palliative-Care-Dienste in der Schweiz - Eine Bestandsaufnahme aus der Perspektive dieser Anbieter	X	X	X
Report (2012)	Bundesamt für Gesundheit BAG, Schweizerische Konferenz der kantonalen Gesundheitsdirektorinnen und -direktoren GDK	Stand und Umsetzung von Palliative Care in den Kantonen Ende 2011			X
Report (2013)	Bundesamt für Gesundheit BAG, Schweizerische Konferenz der kantonalen Gesundheitsdirektorinnen und -direktoren GDK; Wyss N., Coppex P.	Stand und Umsetzung von Palliative Care in den Kantonen 2013			X

Chapter 5: Synthesis and Discussion

This final chapter of the dissertation synthesises and discusses the results of the studies presented in **Chapters 2** to **4**. The conceptual *Model of Caregiver Burden in Parents of Children with LLCs* proposed in **Chapter 1** provided the framework for integrating the findings of the separate studies. This includes a discussion of the sources and determinants of financial burden, that is, a child's LLC, LLC-related events, risk and protective factors. Regarding the latter, the chapter continues with a discussion of how SPPC programmes can assess and address financial burden and, in the Swiss context, how current funding obstacles may impede their efforts. This is followed by a brief discussion of contextual factors. Lastly, strengths and limitations are detailed, and implications for research, policy and practice are suggested. **Chapter 5.7** concludes this dissertation.

5.1 Key findings

This dissertation investigated and explored economic aspects related to families of children with LLCs. It aimed to provide insights into the quantification and measurement of financial burden, to explore hospitalisation and death-related financial and employment implications for parents and to assess the funding landscape of SPPC programmes in Switzerland.

The scoping review presented in **Chapter 2** revealed that existing studies measuring financial burden in families of children with LLCs show little consistency or standardisation concerning applied cost indicators and outcome measures. The review retrieved a wide variety of cost measures that were found to fit into three broad categories: direct costs, indirect costs and financial support. While direct costs included a wide range of OOP medical and non-medical expenses, indirect costs were commonly measured by estimating work-related income loss from illness-related employment disruptions. Financial support was rarely assessed in included studies.

In **Chapter 3** it was shown that families of children with LLCs incur higher travel and accommodation expenses during their child's hospitalisation than during non-hospitalised periods. Although these extra expenses were not very high in the sample analysed, they further added to families' incurred costs. The study furthermore revealed that mothers of children with LLCs were less likely to engage in employment, compared with mothers in the general population. Consistently, 50% or fewer families received financial support. Another key finding of the study is that over one-fifth of the parents who lost their child while participating in the study increased their work commitments within 120 days of their child's death.

The conceptualisation of direct financial flows and funding arrangements regarding SPPC programmes in Switzerland presented in **Chapter 4** demonstrated that current funding structures are complex and fragmented and that funding is derived from public, private and charitable sources. In addition, the 21 experts who participated in the modified Delphi study identified a total of 23 obstacles to and 29 priorities for funding SPPC programmes sustainably, of which the most urgent/pressing fell into the political and structural category. The study showed that improvements in SPPC-related funding models are needed, ranging from a legislative integration, via a full reimbursement of inpatient care costs in the context of high-deficit cases, to an adequate reimbursement of outpatient care and support.

5.2 Sources and determinants of financial burden

As described in **Chapter 1**, families of children with LLCs are at an increased risk of profound financial burden.[1-4] Various sources and determinates may contribute to this increased risk, including primary events related to a child's LLC. In this dissertation, the financial and employment implications of two such events were investigated—a child's hospitalisation and death. In addition, risk and protective factors on the parent and family level may also be of influence.

5.2.1 LLCs and LLC-related events

Generally, every LLC in a child may come with certain financial and employment implications for parents. In **Chapter 2**, a broad range of potential costs were identified. However, as previous studies indicate, the extent of implication may vary depending on a child's diagnosis and demographic characteristics. For instance, studies in paediatric oncology suggest that employment-related income loss is more pronounced in parents of younger children due to the associated higher caregiving demands.[7,8] Moreover, a recent study about rare diseases in children found that families of children whose illness had a shorter duration were more likely to experience overwhelming health expenditures, that is, health spending that exceeded 10% of their total household expenditures or income.[9] Aditionally, families may incur increased costs due to their child's functional limitations and comorbidities. Previous research showed that in children with cerebral palsy, the costs incurred by families were higher when the equipment and technology needs of the child were greater or further devises and equipment were needed as the illness progressed.[3] In studies on children with cerebral palsy[4,5] and congenital heart disease[6] comorbid illness was positively associated with families' OOP medical expenses.

In addition, a family's financial burden may be exacerbated by LLC-related events that parents have to navigate throughout their child's course of illness. For instance, in **Chapter 3** it was shown that families incur higher travel and accommodation expenses during their child's hospitalisation than during non-hospitalised periods. Even though the extra costs were not very high in this study's setting, they further exacerbated their incurred costs. For families whose children undergo extended or recurrent hospitalisations, these expenses can accumulate, further intensifying their financial burden. In addition, in healthcare systems where families lack sufficient insurance coverage, a child's hospitalisations likely result in substantial additional

financial burden for families due to high OOP medical expenses. Evidence from the United States indicates that uninsured children face considerably higher medical costs when compared to their insured counterparts.[10] In Switzerland, families are largely protected from OOP medical expenses due to compulsory health insurance (see discussion of contextual factors below).[11]

Although this dissertation's analyses did not reveal evidence that the death of a child increases families' financial burden via adverse parental employment implications, the time taken off during their child's course of illness likely negatively affects parents' long-term career opportunities and wage development.[7,12] In addition, it has been suggested that funeral expenses constitute a major expense after a child's death,[13] further contributing to a family's financial burden.

Overall, studies comparing families of children with LLCs to those without such conditions (including healthy children) indicate that families of children with LLCs are at an increased risk of financial burden.[7,12,14,15] However, even when comparing families of children with LLCs to those with other chronic, but non-life-limiting, conditions, it becomes apparent that affected families tend to bear a greater financial burden. Studies comparing these two groups indicate that families of children with LLCs incur higher OOP expenses.[4,5,16] Moreover, due to the increased likelihood of prolonged hospitalisation in children with LLCs, the associated costs for their families may also exceed those incurred by families of children with other chronic conditions.

5.2.2 Risk factors

While certain child diagnostic and demographic characteristics as well as LLC-related events, such as a child's hospitalisations, may result in increased costs for families, there may also be risk factors on the parent and family level that place families at an increased risk of financial burden. For instance, as indicated in **Chapter 3** and supported by previous findings,[17-19] families living at a further distance from the hospital are more likely to face increased travel and accommodation expenses.

Additionally, prior investigations have identified three parental characteristics that may place parents at an increased risk of financial burden. This includes single parenthood, being at a younger age and being a mother.[17-20] Both parents of a younger age and single parents are more likely to be in a less robust financial and employment situation.[17-20] Moreover, single parents

may have greater difficulties maintaining their employment due to limitations in sharing caregiving tasks and responsibilities.[18,20] Previous research,[1] along with the findings discussed in **Chapter 3**, highlights that mothers are disproportionately at risk of illness-related employment disruptions. Overall, single parents, parents of a younger age and mothers may be particularly susceptible to financial burden.

Further risk factors include barriers in communicating in the country's primary language (e.g., migrants), a low socio-economic status and other children in the home.[1] Moreover, and considering that a child's LLC can negatively affect family members' health (e.g., back pain and depression),[1,21] health problems in family members may constitute another risk factor. For example, parents may have to give up employment because of negative LLC-related impacts on their health. These families may not only be at risk of financial burden because of their child's illness-related costs but also regarding their own.

5.2.3 Protective factors

In contrast to the risk factors increasing families' likelihood of experiencing financial burden, protective factors may mitigate LLC-related financial adversities and enhance families' resilience against financial burden. One protective factor may be a family's social support network. Previous research suggests that strong social support networks help families to navigate the demanding LLC-related caregiving responsibilities.[22] For instance, friends or relatives may assume household chores and caregiving tasks, such as taking care of siblings or providing care to the child with an LLC.[22] As a result, parents may not have to employ a formal caregiver and may be able to continue in their jobs for longer. Nevertheless, where caregiving responsibilities are assumed by friends or extended family members, the financial burden may simply be shifted onto these informal caregivers, in the form of opportunity costs (**Chapter 2**).

Another protective factor is a family's available financial resources.[17,18,23] In **Chapter 3**, child hospitalisation was no longer associated with travel and accommodation expenses for families with higher incomes. Furthermore, families in high income categories may experience fewer illness-related employment implications. Previous research found that parents with a higher income are less likely to suffer negative LLC-related effects on their employment.[7,18,24] This is possibly the case because they tend to work in positions that are more flexible and provide an option to work remotely.[7,24-26] On the contrary, families with a lower income or less financial

resources may have little financial protection against the adverse financial and employment implications that come with parenting a child with an LLC.

Timely financial support provided early in a child's course of illness may constitute another factor shielding families from financial burden. As noted in **Chapter 2**, financial support can be provided by government and non-government organisations. This may include disability benefits, instrumental support (e.g., food and parking vouchers), income compensation allowances, carers' allowances, assistive equipment and technology grants and other forms of financial assistance.[27] Moreover, affected families have been found to use fundraising efforts to raise additional financial resources to pay for illness-related costs.[28] However, not all families in need may receive financial support. For instance, in the study presented in **Chapter 3**, consistently, 50% or more families did not receive financial support. There may be several reasons explaining why families do not receive financial support: First, due to sufficient financial resources, families may not be in need or eligible for financial aid. Second, financial support may simply not be available. A study about financial support provided to families of children with long-term illnesses conducted in Finland showed that financial support offers differed by region.[29] Third, families may experience difficulties in accessing financial support. Prior research indicates that the application processes can be complex and that families require the support of professionals to access financial support.[20]

Due to the profound psychological and emotional burden, parents of children with LLCs may often struggle to seek and apply for financial support. In parents of children with long-term illness, mental fatigue was suggested to be a major barrier.[29] Therefore, it has been recommended that families receive guidance and support in searching and applying for financial assistance.[24] SPPC programmes may be able to provide such guidance and support and, thus, help protect families against financial burden.

5.3 Financial burden and SPPC

SPPC is a holistic approach aimed at improving the quality of life of children with LLCs and their parents.[30,31] Recognising that illness-related financial burden likely negatively impacts both the parents' and their child's quality of life, SPPC programmes can help by actively addressing families' financial needs. As discussed below, this may start by identifying those families in need.

5.3.1 Financial burden assessment

Conducting financial assessments can help identify families of children with LLCs who are at risk of or are already experiencing financial burden. In paediatric oncology, the current body of literature advocates for the assessment of financial burden as a component of holistic care.[32] This information can be utilised to facilitate targeted support for families' financial needs.[32,33] However, it has been noted that such an assessment may not currently be standard practice in all clinical settings.[32,34]

In detecting families with unmet financial support needs, hospitals and SPPC programmes, may conduct financial assessments at time of initial hospitalisation or enrolment into an SPPC programme. This may include a brief assessment of parents' objective and/or subjective financial burden as well as screening for the risk factors discussed above. Thereby, this initial assessment may not be intended to be a comprehensive assessment of a family's financial situation but rather provide a preliminary indication of its financial burden or its risk of experiencing financial burden.

In addition to this initial financial assessment, studies investigating the support needs of parents of children with LLCs emphasise the importance of regular financial assessments.[32,35] A single assessment at initial hospitalisation or involvement of an SPPC team may fall short in detecting families who experience financial burden at a later stage. The longitudinal evidence provided in **Chapter 3**, which demonstrates that families incur exceedingly high OOP non-medical expenses throughout their child's course of illness, supports this appraisal. Moreover, it was recommended to continue financial assessments even after a child's death, as financial burden may persist into bereavement.[32,35] However, further research may be warranted to show in what time intervals such assessments should be conducted.

Additionally, in **Chapter 2** it became evident that multi-item measurement tools for assessing financial burden in families of children with LLCs are lacking. This may constitute a major barrier to financial assessments not only in research but also in practice. The need for developing standardised multi-item measurement tools to assess the illness-related costs incurred by patients and their families has also been recognised by others.[36] The findings of **Chapter 2**, in particular the generated framework for measuring direct costs—OOP medical and non-medical expenses—may facilitate the development of such measurement instruments.

Utilising the findings of **Chapter 2** for the development of a financial assessment instrument in SPPC, consideration should be given to the context and setting in which an assessment is intended to take place as well as to the vulnerable situation of affected families. For instance, as the generated framework is a generic template, some of its 16 defined cost categories may not be applicable with regard to the local context. Developing a financial assessment tool for Switzerland, for example, one may exclude an assessment of OOP medical expenses because families are protected against such expenses owing to compulsory health insurance.[37] However, in healthcare systems where insurance coverage is insufficient, OOP medical expenses may considerably contribute to families' financial burden and, thus, should be included.

Furthermore, given the vulnerable situation of families of children with LLCs, parents may find continuous financial assessments during illness and bereavement burdensome. As a consequence, they may choose not to complete a financial assessment when it is too lengthy and time-consuming. Hence, to manage the time burden on families, it might not always be practical or feasible to assess all of the framework's 16 categories in detail. For example, an assessment of OOP non-medical expenses for utilities, e.g., electricity, may entail substantial extra effort for parents to provide this information. However, not assessing these costs implies that families may have incurred illness-related costs that were not assessed.

In developing context-relevant measurement tools for use in SPPC and ensuring that the time burden is adequately controlled, engaging families of children with LLCs in the development process may be beneficial. In previous research, engaging patients was found to aid researchers in choosing relevant outcomes and yielding higher retention rates of participants.[38] Engaging parents may provide important insights with regards to both the contextual-relevance of cost indicators and outcome measures and the time needed to complete the assessment.

5.3.2 Financial support provision

Once the financial support needs of a family are identified, adequate support to address these needs should be provided. Thereby, depending on potential budgetary limitations (**Chapter 4**), SPPC programmes may utilise different strategies for providing this support. In **Chapter 1**, three financial support models—the financial counselling, advocacy, and navigation models—were presented.[39]

First, in the financial counselling model, a hospital internal or external financial counsellor assists patients in need of financial support.[39] Utilising this model, SPPC programmes can address parents' financial support needs by referring the family to a financial counsellor. Out of the three models, this model may be the least resource intensive for SPPC programmes because they would not provide further support in addressing families' financial needs. The financial support provision would be fully overseen by a financial counsellor. In general paediatrics, such models were developed and tested in Scotland and Australia, where healthcare professionals referred families in need to welfare advice services.[40,41] Consultations with a financial counsellor were found to enhance families financial well-being by improving parents' financial capability and confidence, managing debt and increasing income.[41] However, the financial counsellor model was criticised as a reactive rather than proactive approach.[39] Moreover, referral to an external financial counsellor causes fragmentation in service provision that may be seen as burdensome by affected families. Prior research indicates that the time and mental demands associated with seeking financial support can further deplete already limited mental resources in parents of children with long-term chronic illness.[29]

Second, the financial advocacy model refers to the direct involvement of healthcare professionals in addressing families' financial support needs.[39] Appling this model in SPPC, members of the SPPC team would directly address parents' financial support needs. This may entail the providing of relevant information, helping with application processes or contacting public and private financial aid programmes. However, for healthcare professionals to receive extensive financial assistance training or to familiarise themselves with governmental and non-governmental financial support programmes, benefit schemes or employment regulations often exceeds what is feasible in clinical practice.[39] This is especially the case in SPPC, where funding obstacles may constrain personnel resources and limit the available time for each family (**Chapter 4**).

Third, in the financial navigation model, a dedicated professional proactively addresses the patient's financial needs, while at the same time stays informed about the patient's medical care and treatment.[39] In the context of SPPC, a dedicated team member may assume this role, for example, a social worker specialised in PPC. This social worker could directly and proactively support families in dealing with the financial and employment implications of parenting a child with an LLC. For instance, one of the key findings of the study presented in **Chapter 3** was that families incur extra travel and accommodation expenses during their child's

hospitalisation. In the financial navigation model, a social worker could proactively address these extra costs by organising parking and food vouchers or free hospital accommodation for parents before a planned hospitalisation. A survey of parents of children with cancer showed that they experienced a social worker's involvement as helpful.[42] Parents valued both the instrumental and logistical support, such as receiving relevant information, help with parking or food vouchers and support in managing work-related matters.[42]

However, having a dedicated professional as a financial navigator in an SPPC team requires considerable financial resources. Ideally, the support provided by this professional would be fully covered within the established reimbursement system. Using the Swiss context as an example, the next subchapter will discuss how SPPC-related funding obstacles may impede the provision of support aimed at addressing the financial support needs of families of children with LLCs.

5.3.3 SPPC funding and financial support provision: A Swiss perspective

Funding issues have been identified to be a major barrier to making SPPC accessible to children with LLCs and their families.[43,44] In Switzerland, access to SPPC remains restricted in many parts of the country.[45] As indicated in **Chapter 4**, this is likely due, in part, to a wide range of funding obstacles that pose a major challenge for the development, implementation and provision of SPPC.

Many of the most pressing obstacles encountered in the current funding of SPPC are part of the political and structural category as indicated by the consulted expert panel (**Chapter 4**). These obstacles range from a lack of guaranteed funding to develop and implement SPPC programmes, via a lack of legal definition of palliative care in general, to an absence of a holistic health policy approach for improving existing funding structures. While these obstacles may constitute a major barrier to the provision of SPPC, including services aimed at addressing families' financial needs, further limitations may arise from the reimbursement tariff-structure-related funding obstacles. On one hand, the absence of tariffs to reimburse certain SPPC services, such as case management, care coordination and support for relatives, likely prevents and restricts service provision to family members. On the other hand, compensation and billing limitations, such as consultation time limits, likely restrict the quantity of support that families are eligible to receive.

As a consequence of these obstacles, SPPC programmes' ability to comprehensively and effectively address parents' financial support needs may be considerably constrained. Budgetary constraints likely limit personnel resources, team members' available time for support delivery as well as the available financial resources for education and training. First, due to funding-related constraints in personnel resources, SPPC programmes may have difficulties employing a designated team member, such as a social worker, who comprehensively addresses the financial support of affected families. Second, limitations in personnel resources may restrict the available time that can be allocated to each patient and family, requiring SPPC teams to set priorities in care and support provisions. Consequently, the time devoted to providing financial support may be reduced. For instance, SPPC programmes may refrain from conducting regular financial burden assessments or providing proactive financial support because it exceeds their available capacity. Third, budgetary constraints may limit SPPC programmes' financial resources to educate and train team members with regards to the provision of financial support. However, continuous education and training would be critical in order for professionals to stay informed about the latest financial support and legal and employment-related regulations.

Today, Swiss SPPC programmes often rely on charitable funding and philanthropic contributions to bridge funding gaps (**Chapter 4**). However, depending on charitable funding and philanthropic contributions to cover the costs of personnel resources is hardly a sustainable long-term solution. In **Chapter 4**, a broad range of priorities for the sustainable funding of SPPC programmes are presented. These include the introduction of care coordination and counselling tariffs, the reduction of quantity and time limitations and the cost-covering reimbursement of such services. Ensuring that SPPC programmes are funded adequately is essential to guaranteeing nationwide accessibility and comprehensive provision of care and support to all children and their families in need.

5.4 Contextual factors

As noted in **Chapter 1**, contextual factors such as social security and healthcare systems likely influence the financial burden borne by families of children with LLCs. While social insurances may influence the financial burden faced by families, healthcare financing regulations may impact their access to effective care and support interventions.

Generally, social insurances can shield families from financial adversity. In Switzerland, several social insurances have been established that protect families from financial burden.[46] For instance, all children and their families living in Switzerland are subject to compulsory health insurance.[11] Moreover, children are exempt from paying deductibles, and co-payments are capped at a relatively low level.[11,37] As a result, all children and their families included in the SPhAERA study were largely protected against OOP medical expenses. Another example is paid care leave and carers' allowances. As parents living in Switzerland are currently entitled to 14 weeks of paid care leave and carers' allowances are provided by the Swiss Disability insurance,[46] the illness-related effects on parental employment and income examined in **Chapter 3** may have been mitigated. Nevertheless, as social security systems are country-specific, families of children with LLCs residing in countries with less established and comprehensive social security systems may not be shielded from illness-related financial adversity to the extent to which families living in Switzerland are shielded. As a consequence, they may endure greater expenses or experience more direct adverse employment implications, increasing their financial burden.

Healthcare financing regulations, as investigated in **Chapter 4**, constitute another contextual factor. As discussed in the previous chapter, funding obstacles can be a major challenge in the provision of care and support to families of children with LLCs. They have the potential to result in unmet financial support. Consequently, adjusting funding regulations can serve as a mechanism to enhance support provision for these families.

5.5 Strengths and limitations

In addition to the strengths and limitations that have been discussed in **Chapters 2** to **4**, this chapter discusses the broader, methodological strengths and limitations of this dissertation. A strength of this research was the use of the scoping review methodology to systematically identify cost indictors and outcome measures used to measure financial burden in families of children with LLCs (**Chapter 2**). By including studies of various designs, the scoping review facilitated a comprehensive mapping and synthesis of the current literature. The findings of the scoping review were then used to inform and guide the assessment of families' OOP non-medical expenses and parents' changes in employment and income via the SPhAERA study.

The SPhAERA study is the first study in Switzerland that comprehensively assesses the effectiveness of SPPC. Its major strength is the prospective collection of longitudinal data that allows for the establishment of a sequence of events, following change over time and relating events to particular exposures.[47] Its longitudinal study design facilitated the use of fixed-effects regression models to investigate child hospitalisation and death-related financial and employment implications for parents (**Chapter 3**). The rigorous controlling for time-invariant heterogeneity and confounding through the applied fixed-effects approach constitutes another strength of this dissertation.

The cohort of children with LLCs analysed in the study presented in **Chapter 3** included children of all four categories of LLCs as defined by TfSL.[48] Moreover, the cohort was heterogenous in terms of demographic and diagnostic characteristics, e.g., age and illness duration. Generally, the population of children with LLCs is a diverse population.[49] Despite the fact that the majority of children included in the analysis had a neurological LLC, the sample may well-represent the diversity of the population.

A further strength of the SPhAERA study and this dissertation was the strong stakeholder involvement. First, an interdisciplinary study team closely collaborated in conducting this research. Second, in designing the SPhAERA study and testing the questionnaires, healthcare professionals and affected families were involved. Third, a diverse set of professionals with expertise in the provision and funding of (paediatric) palliative care in Switzerland was consulted, which allowed for a comprehensive identification of funding-related obstacles and priorities (**Chapter 4**).

Nevertheless, there are several limitations to consider in this dissertation, some of which have already been addressed in **Chapters 2** to **4**. Regarding the generalisability of research findings, it is important to recognise that sampling bias cannot be ruled out in the SPhAERA study. Families experiencing a high caregiver burden at the time of recruitment may have been more likely to decline study participation compared to those with higher levels of physical, emotional or economic resources. Similarly, families experiencing a high caregiver burden during their participation in the study may have been more likely to drop out. Furthermore, only families proficient in German or French languages qualified for participation. Therefore, certain sub-groups of the population, for example, migrants, are likely inadequately represented in the sample. Although language barriers could have been prevented by translating the questionnaire

into various other languages, this was not possible due to the large number of additional resources that such translations would have required.

As recruitment for the SPhAERA study was conducted by medical professionals, study enrolment may have been influenced by gatekeeping as they may have shielded families that they considered to be particularly vulnerable from study participation. The level of gatekeeping may have varied among professionals and across study centres. Early engagement of gatekeepers in the research process and sharing clear information have been recommended in order to decrease gatekeeping.[50]

Another limitation of the research presented in **Chapter 3** was the small sample size. Statistical power may have been limited, making it less likely to detect significant effects (Type II error). To increase statistical power, it would have been necessary to recruit a larger sample. This could have been achieved by including more study centres or extending the recruitment phase further. However, due to resource limitations this was not feasible. An alternative approach for assessing illness-related costs would have been to compare families of children with an LLC to those of children without an LLC. However, as the SPhAERA study solely included children with an LLC such an assessment was not possible.

It is also important to acknowledge that the economic-parts of the SPhAERA study questionnaires were self-developed. Although the questions were designed in accordance with the findings of the scoping review, which likely increased content validity, an assessment of reliability and validity was not conducted. Moreover, certain biases cannot be ruled out, such as response or recall bias. However, the use of discrete response categories and primarily monthly assessments may have prevented bias.

Regarding **Chapter 4**, the experts participating in the modified Delphi study were recruited from various regions across Switzerland. Nevertheless, it is worth noting that there were certain regions, such as the canton of Ticino, where there was no participation from experts. Consequently, and despite the fact that national funding regulation apply to all Swiss cantons, certain canton-specific obstacles to and priorities for funding local SPPC programmes may have not been detected. To circumvent this limitation, it would have been necessary to conduct a nationwide survey, which would have required substantial extra time and financial resources.

In addition, the coronavirus disease 2019 pandemic may have influenced the findings presented in **Chapter 3**. Recruitment and data collection of the SPhAERA study was carried out

throughout the pandemic, which may have had an effect on child hospitalisation rates. While children may have been hospitalised because of a (suspected) infection, planned or non-emergency hospitalisations may have been postponed. Moreover, parents' employment situation may have also been affected as they may have experienced more work flexibility (e.g., working from home), affecting potential hospitalisation-related employment implications.

Overall, considering these limitations, the generalisability of this dissertation's findings is limited. Although the findings of the scoping review presented in **Chapter 2** are likely applicable to various settings and contexts, researchers should be cautious when generalising the findings detailed in **Chapters 3** and **4**.

5.6 Implications

5.6.1 Implications for research

This dissertation aimed to contribute to existing research on economic aspects regarding both families of children with LLCs and the provision of SPPC in Switzerland. While the research presented in **Chapters 2** to **4** provided new evidence regarding these fields of research, further research expanding upon this dissertation's findings is needed.

First, more efforts are warranted to consistently and precisely assess financial burden in families of children with LLCs. Ideally, financial burden should be assessed within a standardised framework that facilitates the synthesis and comparison of research finding across studies. The findings presented in **Chapter 2**, in particular the framework for assessing OOP expenses, may help to assess financial burden in these families more consistently.

Second, future research should investigate further LLC-related employment implications for parents. While this research did not find an association of child hospitalisation with parents' work commitments, it is possible that other LLC-related aspects explain changes in parental work commitments. For instance, future longitudinal studies could explore whether or not parents are more likely to reduce their work commitments at times of increased non-financial caregiver burden, including physical and psychological/emotional burdens. Another potential approach could be to conduct a retrospective assessment of changes in employment starting at or even before the time of diagnosis, for example, by linking employment and health registry data (e.g., as available in the Nordic countries). Thus, researchers could explore whether changes in employment are more likely to occur at or shortly after the initial diagnosis.

Third, more research exploring parents' reasons for returning to work or increasing their working hours in the early bereavement period is needed. In particular, studies could explore whether economic considerations or a desire to return as a means of coping are explanatory factors. Qualitative research can be a valuable approach for examining this phenomenon, for example, by conducting interviews with parents that lost a child due to an LLC.

Another consideration for future research is to develop, test and implement effective models of financial support provision in SPPC. Currently, evidence on how SPPC programmes can effectively address families' financial support needs is largely absent from the literature. The three models of financial support discussed above provide some first insights into how SPPC programmes provide, or could provide, support regarding the financial burden experienced by parents. Research should assess which type of financial support model is most effective in addressing the needs of these families. Intervention research, comparative-effectiveness research and implementation science provide methods and tools to develop, test and implement support interventions. In this context, it should also be noted that research on social workers in PPC is also still scarce.[51]

Last, efforts are needed to quantify gaps in the funding of SPPC in Switzerland. Given the wide range of funding-related obstacles identified in the modified Delphi study presented in **Chapter 4**, SPPC programmes are likely underfunded. However, the precise financial impact of these obstacles remains unclear. In order to explore gaps in funding, researchers may conduct a costing study, which, in a first step, could estimate the costs of an established SPPC programme. In a second step, the data could be used to estimate the costs of an 'ideal' SPPC programme, one that provides care and support according to national and/or international standards and recommendations. This differentiation is critical as current funding obstacles likely constrain personnel resources as well as the care and support provision of established SPPC programmes. Hence, costing an established SPPC programme likely underestimates the gaps in funding. In a final step, the two cost estimates could then be compared with current funding, which would not only reveal current gaps but also any additional funding needed to achieve optimal care and support.

5.6.2 Implications for policy

This dissertation's findings provide valuable indications for policy makers on enhancing the current funding of SPPC in Switzerland. In the study presented in **Chapter 4**, several priorities

for sustainably funding SPPC programmes are presented that highlight areas where policy action is warranted. These range from a legislative integration of palliative care via a harmonisation of regulations across cantons to the adequate reimbursement of inpatient and outpatient care and support. Regarding the latter, this includes the introducing of counselling, case management and coordination fees in outpatient tariff structures to ensure adequate reimbursement, such as supporting families regarding financial, legal and work-related matters.

In addition, policy makers should strive to implement policies and support programmes that effectively and proactively protect families from the adverse financial and employment effects of parenting a child with an LLC. These policies and support programmes may include measures such as disability benefits, financial aid programmes, paid parental leave, income subsidies and carers' allowances. Thereby, policy makers should ensure that all families in need have equal access to support programmes, independent of their socio-economic status and/or socio-cultural background. One way is to raise awareness of available support options and simplify the application processes of governmental support programmes.

5.6.3 Implications for practice

This dissertation's discussion of how SPPC programmes can address parents' financial support needs highlighted two aspects: financial burden assessments and financial support provision. First, conducting financial assessments is essential for the early detection of financial burden in families of children with LLCs. It enables SPPC programmes to initiate proactive support in a timely manner. Given that the findings presented in **Chapter 3** showed that some parents endure substantial cost and/or face profound employment implications throughout their child's course of illness, SPPC programmes should consider conducting such assessments not only at the time of the initial hospitalisation or the SPPC team's involvement but regularly.

Additionally, SPPC programmes should prioritise the establishment of clear strategies (models), processes and guidelines for addressing families' financial support needs. In this dissertation three models of financial support provision were discussed that offer insights into potential approaches for addressing the need for financial support. Depending on available financial and personnel resources and the hospital environment, such as the availability of a hospital-based financial counsellor, SPPC programmes may consider utilising one such model. However, it should be noted that evidence of these models' effectiveness is scarce and further efforts are

needed to determine the most (cost-)effective approach for addressing the financial support needs of families of children with LLCs.

5.7 Conclusions

In addition to the provision of medical and psychosocial care and support, SPPC programmes can support families of children with LLCs by addressing their financial burden. As families' financial support needs vary, depending on the sources and multiple determinants of financial burden, a proactive needs-based provision of support is essential. To facilitate SPPC programmes in providing this support, policy makers should ensure that existing funding obstacles are resolved and that these programmes are adequately funded. All stakeholders, from government to non-government organisations, should ensure the availability of adequate financial support and equal access for all families in need.

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Appendix: Specialised Paediatric PAlliativE CaRe: Assessing family, healthcare professionals and health system outcomes in a multisite context of various care settings: SPhAERA study protocol

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Abstract

Background

The number of children and adolescents living with life-limiting conditions and potentially in need for specialised paediatric palliative care (SPPC) is rising. Ideally, a specialised multi-professional team responds to the complex healthcare needs of children and their families. The questions of, how SPPC is beneficial, for whom, and under what circumstances, remain largely unanswered in the current literature. This study's overall target is to evaluate the effectiveness of an SPPC programme in Switzerland with respect to its potential to improve patient-, family-, health professional-, and healthcare-related outcomes.

Methods

This comparative effectiveness study applies a quasi-experimental design exploring the effectiveness of SPPC as a complex intervention at one treatment site in comparison with routine care provided in a generalised PPC environment at three comparison sites. As the key goal of palliative care, quality of life—assessed at the level of the patient, the family and the healthcare professional—will be the main outcome of this comparative effectiveness research. Other clinical, service, and economic outcomes will include patient symptom severity and distress, parental grief processes, healthcare resource utilisation and costs, direct and indirect health-related expenditure, place of death, and introduction of SPPC. Data will be mainly collected through questionnaire surveys and chart analysis.

Discussion

The need for SPPC has been demonstrated through numerous epidemiological and observational studies. However, in a healthcare environment focused on curative treatment and struggling with limited resources, the lack of evidence contributes to a lack of acceptance and financing of SPPC which is a major barrier against its sustainability. This study will contribute to current knowledge by reporting individual and child level outcomes at the family level and by collecting detailed contextual information on healthcare provision. We hope that the results of this study can help guiding the expansion and sustainability of SPPC and improve the quality of care for children with life-limiting conditions and their families internationally.

Trial registration

Registered prospectively on ClinicalTrials.gov on January 22, 2020. NCT04236180.

Background

Following ongoing medical advances and improved diagnosis and coding, the number of children and adolescents living with life-limiting conditions (LLCs) is rising drastically.[1] Estimates from the United Kingdom showed a prevalence of 26.7 per 10'000 children aged 0–19 years in 2001/2002, and a prevalence of 66.4 per 10'000 in 2017/2018.[1] Many of these children and adolescents need palliative care (PC) services. A new consensus describes PC as '*the active holistic care of individuals across all ages with serious health related suffering because of severe illness and especially of those near the end of life. It aims to improve the quality of life of patients, their families, and their caregivers.*'[2,p.761]. As a special and highly complex subfield of PC, paediatric PC (PPC) is concerned with the support and involvement of the entire family, and aims to impact the patient, family and health system levels. Ideally, to meet the complex healthcare needs of children and their families, a specialised multi-professional team will be available, i.e. PPC services are offered by healthcare professionals specifically trained and working in PPC in the context of a dedicated programme setting.[3]

Specialised PPC (SPPC) programmes most commonly offer a consultative model of care, i.e. by a specialised multi-professional PPC team that either provides direct patient care and family support that goes beyond the affected child's eventual death or provides support and advice to the child's primary care team in- and outside of the hospital.[4] In such a setup, medical specialists (mainly physicians), e.g., neurologists or oncologists refer their patients and families in need. Referrals are mainly driven by symptom burden and the burden of the child's condition on the entire family. However, the referral practices also depend on personal attitudes and motivation of the referrers from the medical specialties towards SPPC. Furthermore, the referrer's perception of evidence supporting SPPC can be considered as a contextual factor influencing referral practices[5] and it is well recognised that compelling scientific evidence on the effectiveness of SPPC is scarce.[6-8]

State of research

The question of whether PC in general is associated with improved patient and caregiver outcomes has been studied and summarised in a meta-analysis of 43 randomised controlled trials with data on 12'731 adult patients.[9] PC interventions were associated with improvements in patient quality of life (QOL) and symptom burden, however, results regarding caregiver outcomes were inconsistent.[9] In PPC, patient-reported outcome assessments are not normally

feasible due to the patient's age or cognitive state. Therefore, in paediatric research, proxy reports from parents are commonly used.[10] A systematic review including eight observational studies found improved QOL in children and family members, improved symptom control, and a positive impact on place of care and family support.[11] The burdens on family members are substantial. Investigating the impact of chronic health conditions on siblings psychological functioning and well-being, Vermaes et al.[12] noted, that the siblings of children with life-threatening conditions appeared especially prone to psychological problems. A recent scoping review, including 34 studies concluded that the experiences these siblings make, impact the way they cope with stress.[13] We found no studies about the influence of SPPC services on the QOL of these siblings.

For healthcare professionals (HCPs), providing compassionate care for children with LLCs and their families is emotionally demanding. High expectations, lack of support and a sense of inadequate preparation, representing demands from the health care system, can lead to stress in care personnel.[14] One purpose of a consultative SPPC service model is to provide support and advice to each child's/family's primary care team, which might ease the care burden and positively influence the QOL of healthcare professionals not specialised in PPC. In a US study of 314 diverse HCPs, 39% were gaged at risk for compassion fatigue, a construct within professional QOL linked to impaired quality of care provision.[14,15]

More evidence related to improved process outcomes at the service level and healthcare resource utilisation is available regarding children/families receiving SPPC. A recent systematic review including 14 cohort studies and one case series found, that the receipt of PPC was associated with a decrease in intensive care use and high-intensity end-of-life care. Regarding hospital admissions, length of stay in hospital, resuscitation orders, and the proportion of hospital and home deaths results were less conclusive.[16] Evidence on healthcare resource utilisation and costs among children, who had accessed a PPC programme versus those, who had not, was summarised in a systematic review.[17] Children enrolled in PPC had fewer hospital admissions, with most studies also showing shorter hospital stays. Conflicting results arose regarding the proportion of patients who received critical care, and calculations of overall healthcare costs were inconclusive.[17] Lower medical costs among PPC recipients through the reduction of healthcare utilisation, however, were recorded in more recent studies.[18,19]

The effectiveness of SPPC as a complex intervention is potentially influenced by many accompanying factors through mechanisms which are not well understood. Important potential

mediators of the receipt of SPPC, e.g., the family's adaptability, should be considered. Based on McCubbin & McCubbin's resilience model of family stress, adjustment and adaptation, a positive relationship has been described between the family's adaptation and the family system, i.e., coherence and family hardiness.[20] Associated with adaptation and considered as central to successful coping with family stressors is the construct of sense of coherence. This refers to an orientation between family members, that makes their reactions to internal and external stimuli structured and predictable, providing resistance resources for handling stimuli, and fostering a sense that life's challenges are meaningful.[21] Family hardiness has been described as a family resource and acknowledged as a mediator in the relationship between stress and illness. As such it has also been related to family members' health and QOL.[22]

Rationale

As more and more countries have recognised the need for SPPC at the policy level, the international development and implementation of PPC programmes have skyrocketed. However, many of those programmes struggle with the transition into routine in- and outside hospital care and therefore sustainability.[4] The question, of how SPPC is beneficial for whom and under what circumstances, remains largely unanswered as validation of innovative care programmes in controlled studies is lacking.[18] Determining clinical effectiveness will require prospective studies using comparison groups to establish the relationship between the receipt of SPPC and selected outcomes on the client and service levels.[23] Additionally, the question of when to initiate SPPC, and the outcomes of early vs. late introduction of SPPC have never been compared, a comparison of the two has been judged a priority in PPC research.[24] Reporting on outcome metrics at the family level and addressing contextual information on healthcare provision has the potential to guide the expansion and sustainability of services and improve the quality of care for children with LLCs and their families internationally.

Objectives

The SPhAERA study's overall target is to evaluate the effectiveness of SPPC and to report on its potential to improve patient-, family-, health professional-, and healthcare-related outcomes. Specifically,

- to explore how SPPC influences the QOL of patients (including their symptom severity and distress), parents and siblings (including grief processes);
- to explore how the availability of an SPPC team influences the QOL of healthcare professionals not specialised in PPC;

- to determine whether the provision of SPPC reduces the utilisation of healthcare resources and costs;
- to determine whether the provision of SPPC reduces direct and indirect health-related costs for families; and
- to evaluate the introduction of SPPC and validation of the Paediatric Palliative Screening Scale (PaPaS-Scale).[25]

Methods

Design and setting

This study applies a quasi-experimental design within the framework of comparative effectiveness research, exploring the effectiveness of SPPC as a complex intervention in comparison with routine care provided in a non-specialised PPC environment. The study takes place at four study sites: the largest Swiss University Children's Hospital with a longstanding established dedicated SPPC programme serves as the intervention site, two other Swiss university children's hospitals and a cantonal children's hospital providing general PPC and are just developing SPPC comprise the comparison sites. Recruitment took place between November 2019 and May 2022, and the longitudinal data collection is ongoing until May 2023. An overview of the study's setup, timeline and outcomes is provided in Figure 1.

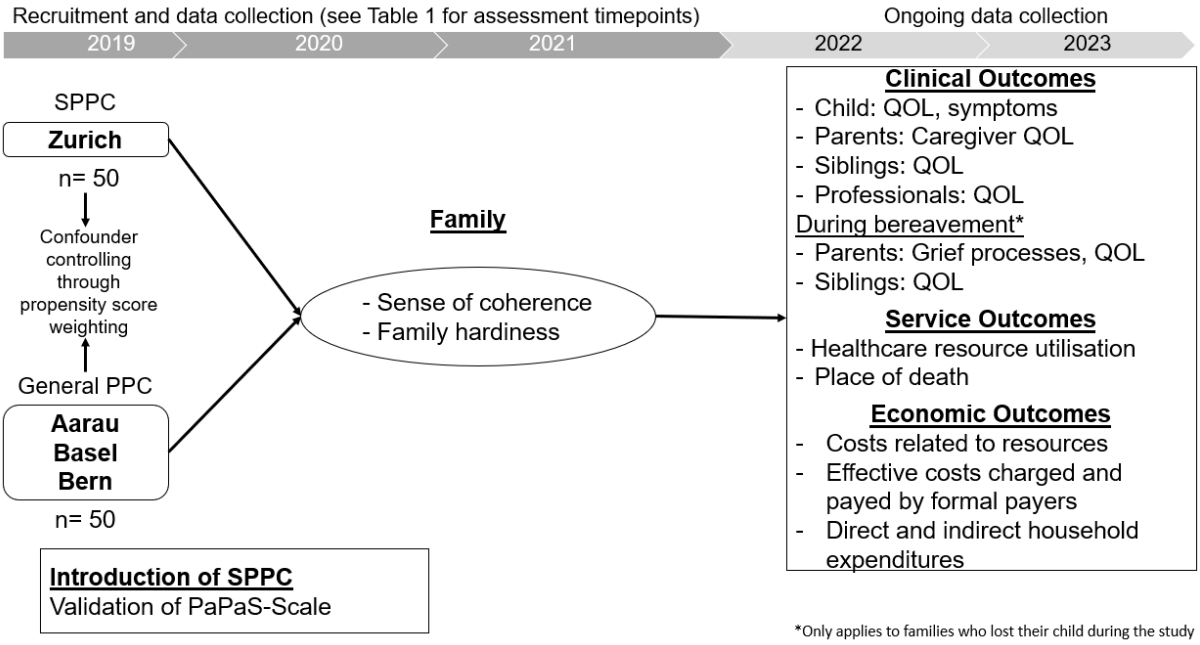

Figure 1: Overview of the study's setup, timeline and outcomes.

Participants and recruitment

Patients and families

Children suffering from an LLC and potentially in need of SPPC, their parents and siblings, as applicable, were eligible to enter the study under the following inclusion criteria: 1) children, aged 0–18 years; parents (mothers and/or fathers) of included children; 2) siblings, aged 8–18 years, of included families; 3) proficiency in German or French language. Neonates with medical complications due to prematurity and/or birth complications and treated in a neonatal intensive care unit and patients enrolled in the SPPC programme with an expected life expectancy of <48 hours were excluded due to limited exposure time. Additionally, children and their families under child protection regulations were not eligible.

For the intervention site, all children entering the SPPC programme, after referral by a member of the frontline care team (usually a physician), were prospectively and consecutively screened for eligibility and invited for study participation. Recruitment was then performed by a member of the SPPC team within the first two weeks of SPPC initiation.

For the comparison sites, recruitment started in February 2020 and was performed by specialists of various medical disciplines, e.g., neurology or oncology after evaluating the potential need for SPPC of their patients. This need was defined per indication criteria of the SPPC programme at the intervention site and read as follows: 1) increase in (unplanned) hospital admissions during the last months; 2) adverse medical events from which the child is not recovering completely; 3) increasing symptom burden; unsatisfactory response to treatments; 4) conflicting treatment goals; 5) estimated life expectancy less than 6–12 months; 6) patient's/parents' wish for PC support.[26]

Healthcare professionals

All HCPs of the following professions working at the study sites or in collaboration with them and involved in the care of the population under study were invited for study participation: physicians, registered nurses, health care assistants, psychologists, social workers, physical therapists, occupational therapists, nutritionists, pastoral workers, logo therapists, social pedagogues, remedial teachers and certified social care workers. To ensure their involvement in PPC, the following inclusion criteria were defined: 1) having cared for patients and their families in the PC phase during a minimum of ten shifts (day/late-shift) or ten consultations and/or 2) having cared for a minimum of two patients in the end-of-life phase and their families or

after the death of the child; 3) employment in their institution for a minimum of three months. Members of the SPPC team at the intervention site were excluded due to their specialisation and dedicated SPPC activities including intervention provision.

Recruitment took place in two cycles: first cycle beginning of 2021 and second cycle beginning of 2022. All HCPs were invited via the written study information which they received through a local coordinator at their institution. The HCPs were instructed to autonomously check their eligibility for study participation based on the criteria listed above.

Intervention and comparison

The service of an SPPC team is considered as a complex intervention with components on various levels and of variable dosing, based on the needs of each individual patient and her/his family. All services provided to patients/families by a member of the multi-professional SPPC team at the University Children's Hospital in Zurich are considered as study intervention. This includes direct medical, nursing, social, and psychological and spiritual consultations of the patient/family, as well as patient-/family-related consultation of the frontline care team. Bereavement support, as integrated part of SPPC is routinely offered at the individual or group level as appropriate for parents and siblings. The programme at the intervention site offers full 24/7 services from the SPPC team's physicians, nurses and psychologists and includes home visits.

For patients/families in the comparison group (Aarau, Basel, Berne) routine care is provided as per established paediatric standards in a generalised PPC environment (provided by disease specific specialists with some PPC training[3]) already in place at all study sites. In all of the three comparison sites a PPC team is available since 2019/2020. However, these developing teams differ on the level of capacity, i.e. few human resources and mostly professionals with basic training and experience in PPC without being fully engaged in PPC, no 24/7 coverage, and limited psychologist and bereavement support.

Assignment to study groups will happen naturally, determined through the recruiting study sites. As this study is conducted in a natural environment with continuous development of care practices, adaptations of care structures and processes at all study sites are possible and probable during the duration of the study. No restraints of this natural evolution will be superimposed

by the study. Care context is assessed on yearly basis in all four participating study sites, described and used as instrumental variables estimation as applicable.

Outcomes and procedures

Improving and maintaining QOL is the core intention of PC and is the main outcome of this comparative effectiveness study. For an overview of the study's setup and timeline, outcomes and measurements, and assessment timepoints see Figure 1 and Tables 1 and 2.

Table 1: Overview of outcomes, measurements and data collection time points.

Outcomes	Data source / Instrument	Domains / Dimensions	Data Collection
Clinical outcomes:			
Primary outcome:			
Caregiver QOL	Quality of Life in Life Threatening Illness–Family Carer Version (QOLLTI-F): 16 items[27]	State of carer, patient wellbeing, quality of care, outlook, environment, finances, and relationships	Baseline at study entry and dynamically after that. Self-report. Mothers, fathers
Secondary outcomes during the child's PC phase:			
Child's QOL	DISABKIDS Chronic Generic Measure - DCGM-37: 37 items[34]	Mental, social, and physical, based on the WHO conceptualisation of health-related QOL.	Baseline at study entry and dynamically after that. Self-report (7 years and older) or proxy report (parent).
Child's symptoms	Memorial Symptom Assessment Scale (MSAS): 30 or 8 items (depending on child's age)[35,36]	Prevalence, severity, distress	
Siblings QOL	KIDSSCREEN-27: 27 items[37]	Physical well-being, psychological well-being, autonomy & parent relation, peers & social support, school environment	Baseline at study entry and dynamically after that. Self-report (8 years and older)
Professional's QOL	Professional QOL (ProQOL): 30 items[38]	Compassion satisfaction, compassion fatigue, i.e. burnout and work-related secondary trauma	Cross-sectional. Self-report. HCPs not specialised in PPC but involved in direct patient care of PPC patients.
Secondary outcomes during bereavement:			
Grief processes	Würzburger Trauerinventar (WüTi): 24 items[39]	Acute emotional and cognitive impairments, general personal development/growth, feelings of guilt and self-reproaches, increase of sensitivity/empathy for others, closeness to the late person	1 month after the child's death and three-monthly after that. Self-report. Mothers, fathers
Parental QOL	The WHO Quality of Life (WHOQOL-BREF): 26 items[40]	Physical, psychological, social relationships, environment	
Siblings QOL	KIDSSCREEN-27: 27 items[37]	Physical well-being, psychological well-being, autonomy & parent relation, peers & social support, school environment	1 month after the sibling's death and three-monthly after that. Self-report (8 years and older)
Mediators of SPPC Outcomes:			
Sense of coherence (SOC)	Family Sense of Coherence (FSOC): 26 items[21]	Comprehensibility, manageability, meaningfulness	Cross-sectional at study enrolment. Structured interview. Mothers, fathers

continued on next page

Outcomes	Data source / Instrument	Domains / Dimensions	Data Collection
Family hardiness (FH)	Family Hardiness Index (FHI): 20 items[22]	Commitment (internal strengths and cooperativeness), challenge (resourcefulness and willingness to learn), control (sense of having control over life circumstances)	Cross-sectional at study enrolment. Structured interview. Mothers, fathers
Service Outcomes:			
Healthcare resource utilisation	Routine data	Number of hospital admissions including number of emergency and/or outpatient consultations and number of admissions to a paediatric intensive care unit (PICU), number of resuscitations, number of invasive procedures e.g., surgery and imaging requiring sedation, total length of hospital stay (LOS) per admission, number of days receiving professional community home care services	Chart review, continuously during the child's PC phase.
Place of death	Routine data	PICU, hospital ward, home, and other	Chart review at time of death
Economic outcomes:			
Economic analysis	Routine data	Costs related to healthcare resources utilised Effective costs charged and paid by formal payers	Cost data retrieved from hospitals and formal payers for the time of study participation.
	Household data	Direct and indirect health-related household expenditures: Out of pocket payments, changes in employment status and income, financial support	Baseline at study entry and dynamically after that. Self-report. Mothers, fathers
Introduction of SPPC:			
Introduction of SPPC and Validation of the PaPaS-Scale	Paediatric Palliative Screening Scale (PaPaS-Scale): 11 items[25]	Trajectory of disease and impact on daily activities of the child, expected outcome of treatment directed at the disease and burden of treatment, symptom and problem burden, preferences/needs of patient or parents / preferences of HCPs, estimated life expectancy	Cross-sectional for each family and child older than one year of age at study entry.
HCPs Outcomes:			
Professional Quality of Life	ProQOL Version 5: 30 items[41]	Compassion fatigue, i.e., burnout and secondary traumatic stress, and compassion satisfaction	Cross-sectional. Self-report. HCPs not specialised in PPC and involved in direct care of PPC or end-of-life patients.

SPPC indicates specialised paediatric palliative care; PC, palliative care, HCP, healthcare professional.

Table 2: Study visits and assessments.

Study Periods	**Care Timepoint**									**Bereavement Timepoint**				
Timepoint[a]	**Screening/ Baseline**	**CT1**	**CT2**	**CT3**	**CT4**	**CT5**	**CT6**	**CT7**	**CT8**	**Death of child**	**BT1**	**BT2**	**BT3**	**BT4**
Time interval in days (Reference = 0)	**0**	**15**	**30**	**60**	**90**	**120**	**150**	**240**	**330**	**0**	**30**	**120**	**210**	**300**
Informed Consent / Demographic Data	x													
Inclusion / Exclusion Criteria	x													
Introduction of SPPC (PaPaS-Scale)	x													
Sense of coherence (FSOC)	x													
Family hardiness (FHI)	x													
Caregiver QOL (QOLLTI-F)	x	x	x	x	x	x	x	x	x					
Child's QOL (disabkids)	x		x	x	x	x	x	x	x					
Child's symptoms (MSAS)	x	x	x	x	x	x	x	x	x					
Siblings QOL (KIDSSCREEN-27)	x	x	x	x	x	x	x	x	x					
Healthcare resource utilisation and costs		x	x	x	x	x	x	x	x	x				
Direct and indirect health-related expenditure	x		x	x	x	x	x	x	x			x		x
Professional's QOL (ProQOL)									x[b]					
Place of death										x				
Grief processes (WüTi)											x	x	x	x
Parental QOL (WHOQOL-BREF)											x	x	x	x
Siblings QOL (KIDSSCREEN-27)											x	x	x	x

SPPC indicates specialised paediatric palliative care; QOL, quality of life.

[a] The timeframe to complete the questionnaire is one week.

[b] Professional's QOL will be assessed after the first year of recruitment and after the second year, i.e., end of recruitment phase.

Patients and families

Since a large proportion of the study population is young or cognitively impaired, we defined caregiver QOL as the primary outcome. Each participating patient/family is followed for a maximum time of one year as long as the child is alive. Assessment timepoints are dynamic, starting with two bi-weekly assessments, followed by four monthly assessments and three-monthly after that. For families, who lose their child during the study, bereavement follow-up continues for another year with four assessments. All outcomes on the family level are assessed as surveys with self-reported and validated questionnaires on paper. Questionnaires are distributed in the hospital or sent home by the study team in case the child is at home at the time of a study assessment. Outcomes on the patient level are assessed as proxy measures through the parents, unless the patient is capable to report her-/himself. Siblings' QOL is only assessed, if they are able to self-report.

Service outcomes are assessed through the collection of routine data via chart review at assessment timepoints. This includes patient-specific information related to healthcare resource utilisation, e.g., hospital admissions and length of stays, procedures, and diagnostic information and date of death. Economic outcomes are assessed through queries for each patient for costs on the healthcare system level (hospitals, formal payers) and through self-reported direct and indirect health-related expenditure data (questionnaire) from participating families.

HCPs

HCPs' professional QOL was retrospectively assessed for one year back after the first and second study year. All HCPs who returned their informed consent received the questionnaire as a hard copy through institutional or postal mail.

All study participants are withdrawn from the study in the case of consent withdrawal or relevant non-adherence to study procedures, i.e. failure to complete questionnaires. No specific follow-up beyond the date of withdrawal/discontinuation is performed and no more data is collected. All data collected up to study withdrawal/discontinuation will be considered for analysis.

Sample size and statistical analyses

We hypothesise that SPPC positively influences the QOL of the caring parents. The null hypothesis is that QOL does not differ between parents of patients in the SPPC programme and the comparison group. The sample size (number of paediatric patients) was calculated to be able to show a difference of 1 point in the QOLLTI-F score[27] one month after study inclusion. The calculations are based on an expected mean QOLLTI-F of 5.5 (on a scale from 0–10) and an expected standard deviation of the QOLLTI-F score of 1.7, as reported in Cohen et al.[27] Note that similar values of mean ±SD, i.e., 5.8 ±1, were observed by Groh, et al.[28]

Sample size was calculated using a resampling method. Each sample size, n i=1, ..., 41=10, ..., 250, was evaluated by simulating R=499 times the QOLLTI-F of n_i individual pairs of caregiving parents from a multivariate normal distribution, using a correlation of 0.8 between parents of the same patient. For each sample size n_i, half of the patients were allocated to the SPPC group and comparison group, with mean QOLLTI-F of 6 and 5, respectively, and a (within group) standard deviation of 1.7. Assuming that only one parent participates in the study for 30% of the patients, this proportion of simulated QOLLTI-F scores for the second parent was omitted, resulting in simulated QOLLTI-F scores for a pair of parents for 70% of the patients, and for only one parent for the remaining 30% of patients. The difference in QOLLTI-F between parents of SPPC vs. comparison patients was then estimated with a linear mixed-effects model with group (SPPC vs. comparison) as fixed factor, modelling a random intercept per patient (to account for the non-independence of parents from the same patient). Sample size was set to ensure at least a power, $1 - \beta$, of 0.8 at a significance level, α, of 0.05.

For this study, a total of 98 paediatric patients should be recruited (i.e., 49 from the SPPC programme and 49 for comparison) to ensure a total of 88 evaluable pairs of caregiving parents, considering a drop-out rate (i.e., proportion of patients who died or withdrew informed consent before QOLLTI-F could be assessed at least once) of 10%. Sample size was estimated using R (Version 3.5.0),[29] using the R packages nlme[30] and sse.[31]

Primary and secondary analyses

The primary outcome QOLLTI-F of the caregiving parents one month after study inclusion will be analysed by a linear mixed-effects model with QOLLTI-F at baseline and group (SPPC vs. comparison) as fixed effects. A random intercept will be modelled per patient (to account for the non-independence of parents from the same patient). In addition, all measurements of

QOLLTI-F taken at different follow-up visits will be analysed by a linear mixed-effects model as above, but with an additional random intercept per parent (to additionally account for the non-independence of multiple measurements per parent).

To adjust the effect size estimate for SPPC vs. comparison for potential confounders, we will use propensity score weighting. Potential confounders were already identified and include characteristics of the patient (diagnosis, age, sex), disease duration, family system (sense of coherence, family hardiness), sociodemographic variables of the family (e.g., parental living situation, family income), and contextual information (e.g., study site characteristics). As a sensitivity analysis, we will add the most important confounders as covariates in the statistical models (analysis of covariance approach).

Secondary endpoints will be analysed by linear mixed-effects models or generalised linear mixed-effects models (in case of non-normal error distribution). Normal linear models or generalised linear models may be used for secondary outcomes measured only once per patient. Sensitivity analyses will be performed as applicable and appropriate.

Missing measurements of the primary outcome, QOLLTI-F at one month, will be multiply imputed, based on measurements taken at two weeks (first follow-up measurement) and available patient characteristics.

Data management, monitoring and risks

The sponsor-investigator is implementing and maintaining quality assurance and quality control systems to ensure that the study is conducted and data are generated, documented (record), and reported in compliance with the protocol, good clinical practice, and applicable regulatory requirement(s). Data extracted from the patient charts will be entered into an internet-based secure data base secuTrial® developed in agreement to the Good Clinical Practice guidelines provided by the Clinical Trials Centre Zurich. Person data will be pseudonymised through secuTrial®. Local coordinators will be assigned to assist with and facilitate logistics concerning the availability of medical charts and workspace in each participating study site. Data collectors will receive instructions and training before the start of data collection to assure and enhance data quality. Furthermore about 5% of the charts will be randomly chosen and data will be collected by two different people for quality checking.

A quality assurance audit/inspection of this study may be conducted by the competent ethics committee. The quality assurance auditor/inspector will have access to all medical records, the investigator's study related files and correspondence, and the informed consent documentation that is relevant to this clinical study. We consider the risk for participants in this study as minimal. Allocation to intervention and comparison groups is determined by the natural environment of the four study sites' care services.

Discussion

This study evaluates the effectiveness of SPPC and its impact on patient-, family-, health professional-, and healthcare-related outcomes. The study will contribute to current knowledge by providing relevant outcome data based on the assessment of SPPC services within a comparative effectiveness research framework. Reporting on outcome metrics at the family level and providing detailed contextual information on healthcare provision has the potential to guide the expansion and sustainability of services and improve the quality of care for children with LLCs and their families internationally.

Determining clinical effectiveness will require prospective studies using comparison groups to establish the relationship between the receipt of SPPC and selected outcomes on the client and service levels.[23] However, the classical randomised controlled trial study design with its origin in establishing the efficacy of new drugs under controlled situations falls short in evaluating the effectiveness of a complex intervention in a real-world setting. Effectiveness research broadens the design options for evaluating interventions by possibly loosening up some of the rigorous control measures mandatory in a randomised controlled trial and therefore trading off strong internal validity in favour of generalisability (external validity).[32] The consequence of less rigorous controls is an increased risk of confounding, which may limit causal interpretation of the study results.

Dealing with potential imbalance between intervention and comparison group will likely be this study's greatest challenge. We will take all possible and reasonable measures, e.g., propensity scoring, multilevel modelling to ensure justified group comparisons and interpretation. The study's rich longitudinal data over a period of approximately one year will allow a unique reporting on courses of illness of the child, of the QOL of close family members and the family's financial burden.

A variety of outcome indicators were used to assess the impact of SPPC so far. Widger et al.[33] found 82 different indicators reported through 46 studies. Among this large number, indicators such as location of death, length of stay in hospital and number of hospital admissions, only 22 indicators were reported in at least two studies.[33] Many of these so far used indicators are also assessed in our study. Others, such as our main outcome of QOL on different levels are less investigated, limiting comparability with our study results.

The main results of this study will be communicated to patients, their family, and the involved healthcare professionals by a letter in lay language. The study group will make every endeavour to publish the data in peer-reviewed journals and we are convinced that this study's comparative effectiveness and longitudinal approach will generate new meaningful evidence to advance the field of PPC internationally.

Ethics approval and consent to participate

The study was approved by the ethics committee Zurich as the leading ethics advisory body (BASEC-Nr. 2019-01170), and by the committees in Nordwest- und Zentralschweiz (for the sites Aarau and Basel), and Berne, and complies with the Swiss Federal act on research involving human beings. Substantial amendments (significant changes) are only implemented after approval of the ethics committees. All children (as applicable) and parents for this study were provided a child/parent information sheet and a child/parent consent form describing this study and providing sufficient information for children and parents to make an informed decision about their participation in this study.

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