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Systematic Literature Review

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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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 3 broad categories: direct costs, indirect costs, and financial support. No study comprehensively assessed all 3 categories. Cost indicators used to measure direct costs were grouped into 5 medical and 11 nonmedical out-of-pocket expenses categories, of which 5 were commonly assessed (ie, 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 article will contribute to a more comprehensive assessment of illness-related financial burden and help guide future policies in this area.

Keywords: cost, family, financial burden, life-limiting, pediatric.

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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 2 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, United Kingdom, prevalence increased from 26.7 to 63.2 (per 10 000) between 2001 to 2002 and 2017 to 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 4 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 nonprogressive 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 hospitalizations, medications and equipment⁷ but often conflicts between employment obligations and childcare responsibilities.⁸

Considerable research has examined families' financial burden in pediatric 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 expenses¹⁰ and 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. Direct costs, that is, out-of-pocket expenses directly incurred by affected families, are commonly divided into medical and nonmedical expenses.¹² Out-of-pocket medical expenses, that is, those directly related to the consumption of healthcare resources, include copayments, deductibles, co-insurances, and all direct charges not covered by formal payers. Out-of-pocket 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 nonmedical. 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 owing to forgone earnings are common indirect costs.¹³ Whether costs are direct or indirect or medical or nonmedical, financial support from both governmental and nongovernmental 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 standardization 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 seem 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 standardization and consistency, we also aimed to categorize 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.

Methods

Study Design

This scoping review was conducted following the 5-step methodological framework (ie, research question identification, study identification, study selection, charting of data, and collating, summarizing, and reporting results) proposed by Arksey

and O'Malley¹⁴ 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.

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

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 (eg, those including nonlife-limiting chronic illnesses such as asthma, allergies, or migraines) were excluded. Studies were also excluded if they assessed perceived rather than economically quantified burden.

Study Selection

Search results were stored and managed via EndNoteX9™ reference software (Clarivate Analytics, London, United Kingdom). After removing duplicates, 2 study team members (T.M. or A.K.G. and S.M.) independently screened titles and abstracts and 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.).

Data Charting and Reporting

We extracted data regarding study characteristics, including study design, participants, assessed outcomes, recall periods, and data collection mode and authors' names, year of publication, and country of origin ([Table 1](#)). Cost indicators and outcome measures were grouped into 3 categories: direct costs, indirect costs, and financial support. Where studies reported direct costs, data were further grouped into out-of-pocket medical and nonmedical 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 categorizations 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

Table 1. Characteristics of included studies.

Author(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 nonmedical	Income loss	Opportunity costs	
Ahuja et al ³² (2019) India	Longitudinal	Families (n = 11) of children (3-18 y) with a diagnosis of cancer	14 wk: 2 wk before diagnosis (questionnaire survey); 12 wk after the diagnosis (cost diary)	X	X			
Badaru et al ³⁶ (2019) Nigeria	Cross-sectional	Families (n = 106) of children (1-11 y) with a diagnosis of cerebral palsy	Per hospital visit or 6 mo 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 y) with a diagnosis of cancer	12 mo (questionnaire survey)			X		
Bona et al ²⁴ (2016) USA	Longitudinal	Families (n = 99 in T1; n = 93 in T2) of children (median = 8.9 y) with a diagnosis of cancer	First interview within 30 days after diagnosis, second interview 6 mo after diagnosis (repeated face- to-face interviews)			X		
Bourke-Taylor et al ²⁹ (2013) Australia	Cross-sectional	Families (n = 29) of children (2-12 y) with a diagnosis of cerebral palsy	Maximum period of 6 y after diagnosis (questionnaire survey)	X	X			
Cohn et al ²⁸ (2003) Australia	Cross-sectional	Parents (n = 100) of pediatric patients with cancer (0.8-18 y)	21-5475 days after diagnosis (questionnaire survey)	X	X	X		
Dockerty et al ⁴³ (2003) New Zealand	Cross-sectional	Parents (n = 237) of children (0-14 y) with a diagnosis of cancer	30 days (questionnaire survey)	X	X	X		X
Dussel et al ²⁶ (2011) USA & Australia	Cross-sectional	American (n = 141) and Australian (n = 89) parents of deceased childhood patients with cancer	Annual income loss assessed at a median of 3.3 y (USA; telephone survey) and 4.4 y (Australia; face- to-face interview and paper-pencil self-report questionnaire) after death			X		

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Table 1. Continued

Author(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 nonmedical	Income loss	Opportunity costs	
Eiser and Upton ⁴⁶ (2006) <i>UK</i>	Cross-sectional	Parents (n = 145) of children (1-20 y) with a diagnosis of any cancer before 16 y of age	Past-wk spending over and above pre-illness expenditure (questionnaire survey)		X			X
Elhoff et al ²³ (2018) <i>USA</i>	Longitudinal; register-based cohort study	Families (n = 481) of children born with severe congenital heart disease	During the first 12 mo of life or until death, if sooner (administrative claims register data)	X				
Fluchel et al ²² (2014) <i>USA</i>	Cross-sectional	Primary caregivers (n = 354) of pediatric patients with cancer (0-18 y)	Per clinic visit (questionnaire survey)		X			
Ghatak et al ³¹ (2016) <i>India</i>	Longitudinal	Families (n = 50) of pediatric patients (1-12 y) with acute lymphoblastic leukemia	During the first mo of therapy, recording expenses on a daily basis (cost diary)	X	X			X
Heath et al ²⁷ (2006) <i>Australia</i>	Cross-sectional	Families (n = 56) of children (mean = 7.6 y) with a diagnosis of cancer	12 mo after diagnosis (questionnaire survey)	X	X	X		X
Hiyoshi et al ⁴⁵ (2018) & Lindahl-Norberg et al ⁴⁴ (2017) <i>Sweden</i>	Longitudinal; register-based cohort study	Families (n = 20 091) of children (0-18 y) with (n = 1899) and without (n = 18 192) cancer	Families were followed for up to 8 y starting from 1 y before the child's diagnosis (register data)			X		
Kamaralzaman et al ⁴² (2018) <i>Malaysia</i>	Cross-sectional	Parents (n = 74) of children (0-18 y) with a diagnosis of cerebral palsy	12 mo (questionnaire survey)	X	X	X		
Lähteenmäki et al ⁴⁰ (2004) <i>Finland</i>	Longitudinal	Families (n = 21) of children (0.1-15 y) with a diagnosis of cancer	First survey 3 mo after diagnosis, second survey 12 mo later (repeated questionnaire survey)	X	X	X		
Mader et al ³⁴ (2020) <i>Denmark</i>	Longitudinal; register-based cohort study	Parents of children with (n = 12 418) and without (n = 125 014) cancer	Follow-up period of 10 y starting 1 y before cancer diagnosis (register data)			X		

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Table 1. Continued

Author(s), Country	Design	Participants	Time horizon and data collection mode	Assessed outcomes				Financial support
				Direct costs		Indirect costs		
				Out-of- pocket medical	Out-of- pocket nonmedical	Income loss	Opportunity costs	
McClung et al ²¹ (2018) USA	Cross-sectional	Families (n = 1956) of children (0-17 y) with congenital heart disease	12 mo (telephone survey)	X				
Michelsen et al ³³ (2015) Denmark	Longitudinal; register-based cohort study	Parents (n = 21 654) of children with (n = 3671) and without (n = 17 983) cerebral palsy	Follow-up period of 28 y, starting 2 y before birth (register data)				X	
Ouyang et al ²⁰ (2012) USA	Cross-sectional	Parents of children (0-17 y) with (n = 112) and without muscular dystrophy	12 mo (telephone survey)	X				
Pagano et al ⁴¹ (2014) Italy	Longitudinal; register-based cohort study	Families (n = 917) of children and adolescents (0- 19 y) with a diagnosis of cancer	3-y follow-up after diagnosis (register data and administrative health data)				X	
Rativa and Carreno ³⁹ (2018) Columbia	Cross-sectional	Families (n = 50) of children (1-14 y) with cancer	Monthly expenses (questionnaire survey)		X			
Sadoh et al ³⁵ (2019) Nigeria	Longitudinal	Families (n = 32) of children (0.5-5 y) with congenital heart disease	Monthly expenses for 3 consecutive mo (repeated questionnaire survey)	X	X			
Schaible et al ¹⁹ (2018) USA	Cross-sectional	Families (n = 744) of children (0-17 y) with cerebral palsy	12 mo (telephone survey)	X				
Sneha et al ³⁰ (2017) India	Cross-sectional	Families (n = 70) of children (mean = 7.8 y) with a diagnosis of acute leukemia	During hospitalization (questionnaire survey and face- to-face interviews)		X		X	
Thomson et al ¹⁸ (2016) USA	Cross-sectional	Families (n = 167) of children (0-18 y) with complex medical conditions	12 mo (questionnaire survey)	X				
Tsimicalis et al ^{37,38} (2012 and 2013) Canada	Longitudinal	Families (n = 99) of children (0-18 y) with a diagnosis of cancer	1 wk per mo for 3 consecutive mo (cost diary and face-to-face interviews)	X	X	X	X	

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Table 1. Continued

Author(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 nonmedical	Income loss	Opportunity costs	
Vessey et al ¹⁷ (2017) USA	Longitudinal	Parents (n = 52) of children (mean = 11.5 y) with a diagnosis of cerebral palsy	During hospitalization (average length of stay = 9.36 days) (cost diary)		X	X		

Results

Study Selection

Our database search yielded 7194 records. After removal of duplicates, screening of titles and abstracts, full-text assessments, and a gray literature search, 30 articles reporting on 28 studies

qualified for inclusion. Figure 1 illustrates the selection process using a PRISMA flow chart.

Characteristics of Included Studies

Of the 28 studies covered by the 30 included articles, 9 were conducted in the United States,^{17–25} with a tenth cross-national

Table 2. Categorized out-of-pocket expenses indicators.

Out-of-pocket medical expenses	Out-of-pocket nonmedical expenses
<p>Treatment and diagnostics 11/16 (69%)*</p> <ul style="list-style-type: none"> - Pharmacotherapy^{23,27,28,31,35-38,40,42,43} (prescription and nonprescription medications) - Therapeutic and surgical procedures³² (eg, surgery, radiation, transfusions) - Special treatments and supportive therapy^{32,36-38,42,43} (eg, chiropractor, massages, physiotherapy, rehabilitation services) - Complementary and alternative therapy^{27,37,38,42} (eg, homeopathy) - Investigations and diagnostic tests^{31,32,36,42} (eg, radiological investigations, laboratory tests) <p>In- and outpatient charges and fees 7/16 (44%)*</p> <ul style="list-style-type: none"> - Inpatient payments and hospital charges^{23,31} - Public hospital bed charges⁴³ - Outpatient charges and payments^{23,43} - Doctor and specialist fees^{28,40,43} - Consultation fees^{36,42} - Ward entrance fees⁴² <p>Equipment and assistive technology 3/16 (19%)*</p> <ul style="list-style-type: none"> - Seating and standing equipment, specialized tables²⁹ - Specialized car, car modifications^{29,37,38} - Mobility devices^{29,36-38} (eg, crutches, wheelchair) - Support equipment^{29,37,38} (eg, eyewear, splints, monitoring devices, ventilator) - Communication devices²⁹ (eg, displays, software) - Equipment for eating and drinking²⁹ (eg, adapted cups, spoons, bottles, tubes) - Equipment for toileting, bathing, and dressing²⁹ - Equipment for sleeping²⁹ (eg, special mattress, body position support, height adjusted bed) <p>Medical aids, dressings, and disposables 3/16 (19%)*</p> <ul style="list-style-type: none"> - Medical aids and dressings^{28,42} - Medical disposables (eg, gloves, syringes)³¹ <p>Other</p> <ul style="list-style-type: none"> - Medical fees and costs for other family members or caregivers as a result of a child's illness³⁶⁻³⁸ <p>Total expenses 4/16 (25%)*</p> <ul style="list-style-type: none"> - Total out-of-pocket medical expenses above and beyond health insurance premiums or reimbursed healthcare costs¹⁸⁻²¹ 	<p>Travel and transport 15/16 (94%)[†]</p> <ul style="list-style-type: none"> - Travel and transport^{17,22,27,28,30-32,35-40,42,43,46} (eg, airfare, gasoline, vehicle maintenance and registration, taxi, parking, public transport, roadway tolls, car rental, mileage) <p>Accommodation 11/16 (69%)[†]</p> <ul style="list-style-type: none"> - Accommodation^{17,22,28,30-32,37-39,42,43,46} (eg, lodging, room or bed rent, rented house, hotel or motel, hospitality house provided by volunteer families, hospital sponsored housing) <p>Food 11/16 (69%)[†]</p> <ul style="list-style-type: none"> - Additional food^{17,27,30-32,36-39,42,43,46} (eg, eating out, snacks, special diet foods during home stay) <p>Childcare and home help 10/16 (63%)[†]</p> <ul style="list-style-type: none"> - Childcare^{17,27-29,36-38,43,46} - Home or domestic help^{17,28,37-39,42,43,46} <p>Communication, internet and cable TV 8/16 (50%)[†]</p> <ul style="list-style-type: none"> - Communication^{28,30,31,37-40,43,46} (frequently recorded: mail, mobile phone, landline costs) - Internet and cable TV³⁹ <p>Clothing 5/16 (31%)[†]</p> <ul style="list-style-type: none"> - Clothing bought because of the child's condition^{28,31,37,38,43,46} <p>Gifts, treats, and toys 4/16 (25%)[†]</p> <ul style="list-style-type: none"> - Gifts or treats for child or other members of family^{37,38,43,46} - Toys or recreational opportunities²⁹ <p>Daily necessities and hygiene products 4/16 (25%)[†]</p> <ul style="list-style-type: none"> - Elements for personal and household hygiene³⁹ - Incidental expenses, daily necessities, supplies (eg, batteries, toiletries)^{17,37,38,42} <p>Utilities 2/16 (13%)[†]</p> <ul style="list-style-type: none"> - Electricity,^{39,43} gas,³⁹ water³⁹ <p>Relocation and home modifications 2/16 (13%)[†]</p> <ul style="list-style-type: none"> - Moving,^{37,38} renovations,^{37,38} home modifications²⁹ <p>Other</p> <ul style="list-style-type: none"> - Special education^{28,42} - Attention to visitors³⁹ - School and extracurricular activities³⁸ - Pet care³⁸ - Funeral expenses⁴³

TV indicates television.

*Number of studies measuring out-of-pocket medical expenses reporting at least one indicator in the respective category.

[†]Number of studies measuring out-of-pocket nonmedical expenses reporting at least one indicator in the respective category.

Table 3. Categorized indirect cost measures.

Income loss	Opportunity costs
<p>Self-report income data 11/15 (73%)*</p> <ul style="list-style-type: none"> - Estimation of income loss by valuing illness-related work disruptions,^{17,24-28,30,37,40,42} for example: <ul style="list-style-type: none"> ■ Cutting back on work hours and commitments ■ Quitting a job ■ Forgone overtime ■ Unpaid leave ■ Closing or suspending business - Estimation of income loss by subtracting parent-reported income before diagnosis from parent-reported income before study participation⁴³ <p>Income data from registers 3/15 (20%)*</p> <ul style="list-style-type: none"> - Estimation of income loss by analyzing 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: <ul style="list-style-type: none"> ■ Total income (all sources of income) ■ Income from work ■ Several types of benefits (eg, unemployment, sickness, childcare related) 	<p>National Census Wage 1/15 (7%)*</p> <ul style="list-style-type: none"> - Informal caregiving time spent by both mothers and fathers valued using US National Census Wage data³⁷ <p>Regional gross domestic product 1/15 (7%)*</p> <ul style="list-style-type: none"> - Informal caregiving time spent by one of the parents valued by the per diem regional gross domestic product⁴¹

*Number of studies measuring indirect costs grouped in the respective category.

study involving both Australian and US participants.²⁶ Additionally, 3 each were conducted in Australia²⁷⁻²⁹ and India,³⁰⁻³² 2 each in Denmark^{33,34} and Nigeria,^{35,36} and 1 each in Canada,^{37,38} Colombia,³⁹ Finland,⁴⁰ Italy,⁴¹ Malaysia,⁴² New Zealand,⁴³ Sweden,^{44,45} and the United Kingdom.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, 7 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 2^{24,40} or 3³⁵ assessment time points. Families were asked to record expenses in cost diaries for a maximum of 12 weeks.³² Three population-based studies, 2 conducted in Denmark and 1 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 pediatric cancer.^{22,24-28,30-32,34,37-41,43-46} and 6 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 al²⁰ surveyed families with children suffering from muscular dystrophy, and Thomson et al¹⁸ examined a sample of children with complex medical conditions, including neurological and congenital conditions and pediatric 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 3 categories—direct costs (out-of-pocket medical and nonmedical), indirect costs (income loss and opportunity costs), and financial support. No study comprehensively assessed all 3 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 2 the opportunity costs of informal caregiving.^{37,41} The extent to which out-of-pocket medical^{18-21,23,27-29,31,32,35-38,40,42,43} and nonmedical^{17,22,27-32,35-40,42,43,46} expenses were measured varied across studies. Financial support was assessed in 4 studies.^{27,31,43,46} All included studies' characteristics are presented in Table 1.

Direct Costs: Out-of-Pocket Medical and Nonmedical Expenses

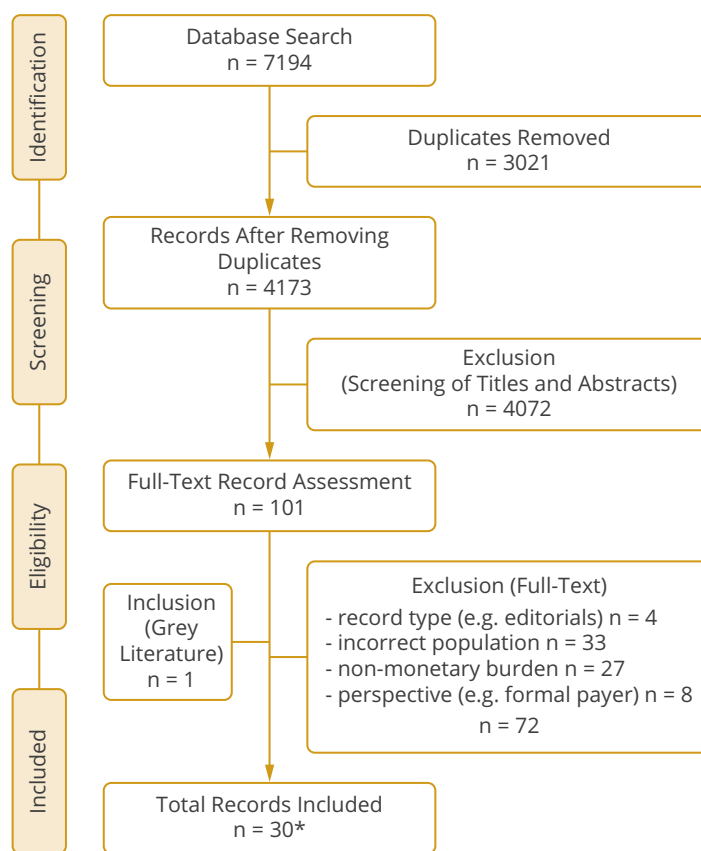
Both out-of-pocket medical and nonmedical expenses were reported for varying periods of time and to different extents.

Table 4. Type and source of financial support.

Type	Source
<p>Assistance with medical expenses 4/28 (14%)*</p> <ul style="list-style-type: none"> - Assistance with treatment expenses and care costs^{27,31,43,46} 	<p>Governmental 4/28 (14%)*</p> <ul style="list-style-type: none"> - Financial support received/available from governmental sources^{27,31,43,46} (eg, disability living allowance, carer's allowance)
<p>Assistance with nonmedical expenses 3/28 (11%)*</p> <ul style="list-style-type: none"> - Assistance with nonmedical expenses^{27,43,46} (eg, transportation, accommodation) 	<p>Nongovernmental 3/28 (11%)*</p> <ul style="list-style-type: none"> - Financial support received from nongovernmental sources^{27,31,46} (eg, charities, support groups, fund-raising efforts)

*Number of total studies reporting financial support within the respective category.

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



*included studies n = 28 (2x two articles reporting on the same study)

Out-of-pocket 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 al²⁹ and Cohn et al²⁸ reported expenses since diagnosis. In the case of out-of-pocket nonmedical expenses, although 5 studies reported monthly figures,^{31,35,36,39,43} others used shorter or longer assessment periods, ranging from daily³⁰ to annual^{27,42} or even multi-year assessments.²⁸

Seventeen studies aggregated individual indicators to estimate total out-of-pocket 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 out-of-pocket medical and 11 out-of-pocket nonmedical). In addition, 4 studies estimated total out-of-pocket 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 out-of-pocket medical expenses, the costs of pharmacotherapy (ie, direct charges, co-payments, and deductibles for prescription and nonprescription 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 out-of-pocket medical expenses, for example, therapeutic and surgical procedure costs, were only assessed within specific contexts. For instance, in India, Ahuja et al³² 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 al²⁹ focused their study on equipment and assistive technology expenses, because 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 al³⁸ assessed expenses for 16 indicators aggregating a total of 74 breakdown items, including specific medications (eg, antibiotics, antiemetics, or antipyretics). In comparison, Ahuja et al³² reported expenses for only 7 indicators. Both studies assessed out-of-pocket expenses incurred by families of children with a diagnosis of cancer.

Common indicators assessed regarding out-of-pocket nonmedical 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 al³⁸ aggregated 11 breakdown items including taxi fares, public transit, parking, and airfare. Other studies measuring out-of-pocket nonmedical expenses provided total travel and transport expenses only, with no itemization.^{17,31,46}

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 2 studies.^{37,41} These valued informal caregiving at market wage rates,^{37,41} but based their calculations on different source data. Tsimicalis et al³⁷ valued caregiving time according to US National Census Wage data; Pagano et al⁴¹ 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 al⁴³ 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.

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 nongovernmental^{27,31,46} sources, such as carer's allowances, charity grants, support groups, or fund-raising efforts. Table 4 provides an overview of the types and sources of financial support provided.

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 3 broad categories: direct costs (out-of-pocket medical and nonmedical expenses), indirect costs (income loss, opportunity costs), and financial support. Although most reviewed studies covered 1 or 2 of these categories, none comprehensively assessed all 3. Reporting gaps were particularly broad regarding incoming financial support and the opportunity costs of informal caregiving.

In addition, studies showed little consistency or standardization concerning applied cost indicators and outcome measures. This is particularly apparent in assessments of out-of-pocket medical and nonmedical 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 (eg, benefits and allowances) are insufficiently recognized. 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 standardization 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) and general recommendations for advancing research on financial burden and enhancing cost-of-illness studies' methodological consistency.

Direct Costs

Considering the pronounced variations in the extent to which out-of-pocket expenses were assessed across studies and the fact that we could not locate any standardized 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 out-of-pocket 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 out-of-pocket expenses. Although a number of indicators (eg, prescription and nonprescription medication) may be applicable across contexts and settings, specific LLCs and healthcare system characteristics may require context and setting-specific indicators. For instance, because 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 out-of-pocket nonmedical 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 al³⁸ 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 and renovation and home modification expenses were major drivers of financial burden.^{28,38} Nevertheless, across the reviewed studies, these costs were infrequently assessed, indicating that direct costs may be commonly underestimated.

To enable more consistent and standardized 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 standardized framework will allow comprehensive, comparable assessments of out-of-pocket expenses and facilitate between-study comparisons. Methodological issues potentially arising with this framework's application are discussed below. Although 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 nonlife-limiting, conditions.

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' socioeconomic situations observed a high prevalence of adverse employment effects, particularly among mothers,⁹ attention to

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*	Treatment and diagnostics include all family-incurred expenses related to procedures to diagnose and treat a disease or injury and improve health and health-related well-being.	<ul style="list-style-type: none"> - Out-of-pocket medical expenses include direct charges, co-payments, deductibles, and co-insurances. - Out-of-pocket nonmedical expenses include additional illness-related expenses above and beyond ordinary family-incurred costs. - Sets of indicators for measuring out-of-pocket medical and nonmedical expenses should be chosen in consideration of context-specific factors (eg, 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 <i>methodological considerations</i>). - Consideration should be given to the research burden affected families might experience.
	In- and outpatient charges and fees*	In- and outpatient charges and fees include all family-incurred charges and fees for in- and outpatient services (eg, hospital bed charges, consultation fees).	
	Equipment and assistive technology*	Equipment and assistive technology include all family-incurred costs related to equipment and supportive technology needs.	
	Medical aids, dressings, and disposables	Medical aids, dressings, and disposables include all occasional and incidental expenses that may be additionally incurred by families (eg, gloves, wound dressings).	
	Other expenses	Other expenses include potential further family-incurred out-of-pocket medical expenses (eg, medical costs for other family members as a result of a child's illness).	
Out-of-pocket nonmedical	Travel and transport	Travel and transport include all family-incurred illness-related travel and transport expenses. [†]	
	Accommodation	Accommodation includes all family-incurred illness-related accommodation expenses. [†]	
	Food	Food includes all family-incurred illness-related expenses for food above and beyond ordinary expenses. [†]	
	Childcare and home help	Childcare and home help include all family-incurred illness-related childcare and home help service expenses.	
	Communication, internet, and cable TV	Communication, internet, and cable TV include 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.	
	Gifts, treats, and toys	Gifts, treats, and toys include 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 include all occasional and incidental expenses that are additionally incurred by families because of a child's illness (eg, batteries, soap).	
	Utilities	Utilities include all family-incurred illness-related expenses for utilities (eg, water, electricity, gas) above and beyond ordinary expenses.	
	Relocation and home modifications	Relocation and home modifications include all family-incurred expenses for relocating or performing renovations and home modifications because of a child's illness.	
	Other expenses*	Other expenses include all other family-incurred illness-related out-of-pocket nonmedical expenses not captured by other categories.	

LLC indicates life-limiting condition; TV, television.

*Potential indicators, see Table 2.

[†]Potential cost items, see Table 2.

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 (eg, number

of reduced working hours, days of unpaid leave), others used general estimates of overall income loss (eg, by using ordinal response categories). We also noted that most studies disregarded the potential mitigating effects of financial coping strategies (eg, 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 (eg, 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 analyzing long-term illness-related effects on parental income beyond employment disruptions.

Regarding the opportunity costs of informal caregiving, traditional assessments involve 2 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

Financial Support

Only 4 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, different types and extent of financial support precluded an overall synthesis. Categorizing financial support according to designated use (eg, assistance with out-of-pocket medical or nonmedical expenses) and source may increase reporting transparency.

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 (eg, 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 more 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 unnecessary 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 3 consecutive months. Validation of self-report data, for example, against official tax records (income loss) or receipts (out-of-pocket expenses), should also be considered to address recall errors.

We observed high variability both in the number of cost items (eg, gasoline, parking tickets, road tolls) aggregated within an indicator (eg, transportation) and in the overall number of included indicators aggregated to determine total out-of-pocket medical and nonmedical 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.

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. Nevertheless, we did not analyze and synthesize 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.

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 3 relevant financial categories: direct costs, indirect costs, and financial support. To help standardize this task, we generated a framework for measuring out-of-pocket 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.

Supplemental Material

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