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A systematic scoping review of the recent literature (~2011–2017) about the costs of illness to parents of children diagnosed with cancer

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ABSTRACT

Purpose: The study purpose was to map and identify gaps in the recent (~2011–2017) literature on the costs of illness to parents of children diagnosed with cancer. The costs of illness include direct costs, indirect costs and psychosocial costs.

Methods: A systematic scoping review was conducted. Data sources included PubMed, CINAHL, PsychInfo and EconLit. Studies were eligible for inclusion if they were conducted in high-income countries, published in the English language, and reported parent perspectives on direct costs, indirect costs and/or psychosocial costs due to financial costs.

Results: 25 studies were eligible. Most were conducted in Canada, the USA, or Sweden. The studies used a variety of designs, target populations, time frames and sample sizes. Intervention studies were lacking. Across studies fathers were underrepresented. While no study comprehensively measured costs of illness, more studies used rigorous methods and considered psychosocial costs. Financial costs were measured using a micro-costing or general estimates approach. Psychosocial costs were measured using a variety of PRO measures, some of which were investigator developed. The studies provide evidence that financial toxicity occurs in pediatric oncology.

Conclusions: Future studies should comprehensively measure costs using a consistent set of established measures and make efforts to recruit fathers to cost of illness research. Interventions to mitigate financial toxicity are needed.

1. Introduction

Childhood cancer is costly to both the healthcare system (Kaul et al., 2015; Price et al., 2009) and to parents (National Academies of Sciences, Enginering & Medicine, 2015). Costs to parents include the direct (out-of-pocket expenditures) and indirect (productivity loss) costs of meeting their child's medical and informal caregiving needs. Together, these direct and indirect costs generate illness-related financial burden, which in turn produces psychosocial (somatic and psychological symptoms, quality of life declines) costs for parents and families (Hodgson and Meiners, 1982). In adult oncology, this problem has been termed "financial toxicity" (de Souza et al., 2014, 2017; Zafar and Abernethy, 2013a, 2013b). Cancer-related financial costs are potentially "toxic" in that, in addition to any psychosocial costs incurred, patients and caregivers may attempt to control cancer-related financial costs by using financial coping behaviors (tangible efforts to ease financial burden). Risky financial coping behaviors include suboptimal adherence to recommendations from health professionals regarding cancer therapy, monitoring for treatment toxicities and disease recurrence, and lifestyle behaviors (regular primary care, healthy dietary intake, regular physical activity) to control risk for co-morbid conditions. Such risky financial coping behaviors can augment existing disparities in cancer-related health outcomes (de Souza et al., 2014). To our knowledge, the term financial toxicity has not been used in the pediatric oncology literature.

To determine the state of knowledge about costs of childhood cancer to parents and families, Tsimicalis et al. (2011) systematically reviewed 13 eligible studies published over a 31-year time period (1979–2010) and evaluated their quality. Their review identified considerable methodological heterogeneity in the included studies, which caused difficulties with the evaluation component of the review and limited the researchers' ability to make across-study comparisons. The review also found that while all included studies identified considerable direct or indirect costs, few assessed both of these cost components or conducted the assessments in methodologically rigorous ways. Further, while two studies included in their review reported negative effects of financial costs on psychosocial aspects of life (vacations, hobbies, social activities), no study assessed psychological costs in detail nor did any

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study include an established measure of quality of life. The authors concluded that the enormity of the costs of childhood cancer to parents and families remains unclear (Tsimicalis et al., 2011).

Over the six years since publication of the Tsimicalis et al. review, direct medical costs (cost to the healthcare system for providing products and services to a patient or patient population) have persistently risen. Simultaneously, as a means to control direct medical costs, care has continually shifted to outpatient settings, thus increasing caregiving burden and the associated costs to caregivers. During the same time period, job security has been evasive, wages stagnant and the cost of living has mounted. In the United States of America (USA) for example, 41% of working-age adults report not having $400 to cover an emergency expense (Board of Governers of the Federal Reserve System, 2017), never mind the exorbitant financial costs associated with the diagnosis, treatment and recovery from childhood cancer. Although the 2010 Patient Protection and Affordable Care Act has increased health insurance coverage in the USA, many working-aged adults and their children are under-insured for catastrophic illnesses like cancer. Adults with lower-incomes struggle to pay health insurance premiums, deductibles, co-pays and prescription drug costs (Obama, 2016). Across high-income countries, undocumented refugees, migrants (De Vito et al., 2015) and immigrants (Hacker et al., 2015) experience multiple barriers to accessing even basic health care for themselves and their children.

During the same time period, assessment of persons' perceptions of their health care experiences, that is, person-reported outcomes (PROs) in research and clinical practice have been recognized as critical to improving health care quality (National Quality Forum, 2013). Although costs of illness to patients, caregivers and families (as compared to direct medical costs) have been difficult to measure in the past (Russell and Bernhardt, 2016; Tsimicalis et al., 2011), digital technologies, recognition of the importance of PROs, and development and validation of PRO measures and item sets now provide means to overcome barriers to collecting these data (Basch and Bennett, 2014). Furthermore, strengthening the rigor of research in the area of "financial toxicity" requires comprehensive measurement of the financial components of the costs of illness (direct costs, indirect costs, financial burden), of the financial coping behaviors that interventions might target and of the consequent psychosocial costs overtime, across the illness trajectory and across populations using rigorous methods and validated measures (Gordon and Chan, 2017; Tsimicalis et al., 2011).

In summary, contemporary parents likely bear greater costs due to childhood cancer than in the past due to the socio-economic environment. Measurement of the costs of childhood cancer to parents and families may have improved since the Tsimicalis and colleagues review given the recognition of the importance of PROs and availability of digital technologies for data collection. Moreover, determining the enormity of the costs of illness using established methods is essential to providing high quality evidence-based comprehensive care for children diagnosed with cancer and their families, and to informing policies to support parents who play vital roles in maximizing medical and quality of life outcomes for children diagnosed with cancer (National Academies of Sciences, Engineering and Medicine, 2015).

The objective of this study is to identify advances and gaps in the recent literature about costs of illness to parents of children diagnosed with cancer and their families. To accomplish this objective, we conducted a systematic scoping review of the recent (~2011–2017) research to address the following research questions:

1. What is the map of this research area?
2. How did recent studies measure the cost of illness components?
3. Do these studies suggest that financial toxicity happens in pediatric oncology?
4. What profile describes parents at risk for financial toxicity?

2. Conceptual framework

Consistent with the Tsimicalis et al. review, we examined costs of illness as conceptualized by Hodgson and Meiners (1982) to include the psychosocial costs generated by direct and indirect costs of illness. *Direct costs* include out-of-pocket expenditures related to the diagnosis and treatment of childhood cancer, to supportive care, to post-treatment monitoring and rehabilitation of cancer-related impairments, to end-of-life care, and to maintaining the family household and household routines while parents attend to the patient's medical needs and provide caregiving. *Indirect costs* include productivity loss due to parental work and/or education disruptions due to caregiving (Hodgson and Meiners, 1982). Indirect costs are reflected in reduction in annual household income and allocation of other assets with monetary value, including time, to address the child's illness (Anderson et al., 2007). *Psychosocial costs*, or declines in quality of life, include new onset or worsened stress and psychological symptoms, poorer psychosocial functioning, deteriorated family function, and degraded living conditions due to illness-related financial burden (Hodgson and Meiners, 1982). Psychosocial costs might also include new onset or worsened somatic symptoms, and poorer lifestyle behaviors (The Family Caregiving Alliance, 2016) attributable to illness-related financial costs and financial coping behaviors.

2.1. Methods

Our review process was guided by the research questions and the steps for conducting systematic scoping reviews as recommended by the Joanna Briggs Institute (Peters et al., 2015). Systematic scoping reviews are well suited to map a research area, clarify concepts and their boundaries, and identify key factors and knowledge gaps to generate recommendations for future research. This type of systematic review is typically undertaken when there are reasons to suspect, as we did, that full synthesis might not be feasible given methodological heterogeneity or dearth of studies that meet the inclusion criteria. Systematic scoping reviews differ from other types of systematic reviews in that evidence from the included papers is summarized, not synthesized. Additionally, formal assessment of the quality of the included studies is not a review component (Arksey and O'Malley, 2005; Levac et al., 2010; Peters et al., 2015; Tricco et al., 2016).

2.2. Inclusion and exclusion criteria

To be included in the review, studies were required to meet the following criteria: (a) the study investigated the costs of any type of childhood cancer from the perspective of parents living in a country with a high-income economy where state-of-the-science medical and supportive care for children diagnosed with cancer is widely available; (b) data were collected regarding direct costs, indirect costs and/or subsequent financial burden to parents; and (c) the financial coping behaviors and psychosocial costs, if any were described, were explicitly tied to childhood cancer-related financial costs or financial burden. The caregiving may have occurred during any phase of the cancer trajectory and in any care setting.

One of our initial assumptions was that, unlike the USA, other countries with high-income economies as defined by the World Bank (World Bank, 2017) have social policies that completely protect their official residents from financial costs of illness. However, screening identified papers by title and abstract suggested otherwise. Thus, otherwise eligible papers describing studies conducted in any country with a high-income economy were included in the review. Although we did not expect studies using qualitative approaches to have "measured" costs of illness, studies that applied quantitative and/or qualitative approaches were eligible for inclusion. From the qualitative studies, we sought guidance about specific psychosocial costs and other relevant concepts that should be measured in future costs of illness studies using