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Costs for Childhood and Adolescent Cancer, 90 Days Prediagnosis and 1 Year Postdiagnosis: A Population-Based Study in Ontario, Canada

Claire de Oliveira, PhD[1,2,3,4], *Karen E. Bremner, BSc*[5,*], *Ning Liu, MSc*[4], *Mark L. Greenberg, MD*[6,7], *Paul C. Nathan, MD, MSc*[2,7], *Mary L. McBride, MSc*[8,9], *Murray D. Krahn, MD, MSc*[2,3,4,5,10,11]

[1]*Centre for Addiction and Mental Health, Toronto, Ontario, Canada;* [2]*Institute of Health Policy, Management and Evaluation, University of Toronto, Toronto, Ontario, Canada;* [3]*Toronto Health Economics and Technology Assessment Collaborative, Toronto, Ontario, Canada;* [4]*Institute for Clinical Evaluative Sciences, Toronto, Ontario, Canada;* [5]*Toronto General Research Institute, University Health Network, Toronto, Ontario, Canada;* [6]*Pediatric Oncology Group of Ontario, Toronto, Ontario, Canada;* [7]*Hospital for Sick Children, Toronto, Ontario, Canada;* [8]*British Columbia Cancer Agency, Vancouver, British Columbia, Canada;* [9]*University of British Columbia, Vancouver, British Columbia, Canada;* [10]*Faculty of Pharmacy, University of Toronto, Toronto, Ontario, Canada;* [11]*Department of Medicine, University of Toronto, Toronto, Ontario, Canada*

ABSTRACT

Background: Childhood and adolescent cancers are uncommon, but they have important economic and health impacts on patients, families, and health care systems. Few studies have measured the economic burden of care for childhood and adolescent cancers. **Objectives:** To estimate costs of cancer care in population-based cohorts of children and adolescents from the public payer perspective. **Methods:** We identified patients with cancer, aged 91 days to 19 years, diagnosed from 1995 to 2009 using cancer registry data, and matched each to three noncancer controls. Using linked administrative health care records, we estimated total and net resource-specific costs (in 2012 Canadian dollars) during 90 days prediagnosis and 1 year postdiagnosis. **Results:** Children (≤14 years old) numbered 4,396: 36% had leukemia, 21% central nervous system tumors, 10% lymphoma, and 33% other cancers. Adolescents (15–19 years old) numbered 2,329: 28.9% had lymphoma. Bone and soft tissue sarcoma, germ cell tumor, and thyroid carcinoma each comprised 12% to 13%. Mean net prediagnosis costs were $5,810 and $1,127 and mean net postdiagnosis costs were $136,413 and $62,326 for children and adolescents, respectively; the highest were for leukemia ($157,764 for children and $172,034 for adolescents). In both cohorts, costs were much higher for patients who died within 1 year of diagnosis. Inpatient hospitalization represented 69% to 74% of postdiagnosis costs. **Conclusions:** Treating children with cancer is costly, more costly than treating adolescents or adults. Substantial survival gains in children mean that treatment may still be very cost-effective. Comprehensive age-specific population-based cost estimates are essential to reliably assess the cost-effectiveness of cancer care for children and adolescents, and measure health system performance.

Keywords: adolescent cancer, Canada, childhood cancer, costs and cost analysis.

Introduction

Children aged 0 to 14 years represent less than 1% of patients newly diagnosed with cancer in North America [1,2], but the burden of cancer on patients and their families is very high. Treatment is resource-intensive and costly for health care systems and families [3–6].

Incidence and survival statistics provide information concerning disease burden and the effects of improved diagnosis and treatments [7,8]. Studies describing the cost of care are analogues of descriptive epidemiological studies. They characterize disease burden as the financial burden borne by patients, families, and health systems. This information is useful for hypothesis generation, and to plan budgets, prioritize research, and report financial indicators for cancer system performance [9,10]. Most importantly, costing studies provide a scientific foundation for robust and reliable inputs for cost-effectiveness studies of cancer treatments [11,12].

Conflicts of interest: The authors have no conflicts of interest. Parts of this material are based on data and information compiled and provided by the Canadian Institute for Health Information as well as by Cancer Care Ontario. Nevertheless, the analyses, conclusions, opinions, and statements expressed herein are those of the authors, and not necessarily of either of these organizations. No endorsement is intended or should be inferred.

* *Address correspondence to:* Karen E. Bremner, Toronto General Hospital, Room EN10-231, 200 Elizabeth Street, Toronto, Ontario, Canada M5G 2C4.

E-mail: kbremner@uhnresearch.ca.

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Most studies of the costs of childhood and adolescent cancer therapy are outdated, use small samples, or include only one type of cancer or health care service [4,13–17]. Consistent methods and standards of reporting are required for the interpretation and practical application of cost estimates [9,10].

Also, existing studies often group patients aged 15 to 19 years with children or older adults. This age group is, however, unique in that embryonal and hematological cancers are less common than in young children, and few patients have tumors typical of adults [2,18,19]. Although centers for adolescents and young adults exist in some areas, these patients often receive care at either a pediatric or an adult facility, frequently receiving different treatment protocols and supportive care depending on the locus of care [20–22].

In Canada, universal health care insurance is publicly funded, mostly through taxation, and provides coverage for almost all medically necessary health care (including the direct medical costs of cancer therapy) for all permanent residents (including children) regardless of medical history or personal income. Each province administers its own insurance plan within guidelines set by the federal government under the Canada Health Act [23]. The Ontario Health Insurance Plan (OHIP) covers all residents of Ontario, Canada's most populous province, under the direction of the Ontario Ministry of Health and Long-Term Care. Private health insurance is not available or required for the medically necessary services covered by OHIP, but can be obtained for services not covered by OHIP, such as most dental care, allied health, and outpatient prescription drugs (covered for only those aged 65 years and older, on social assistance, or with high drug costs relative to their income).

We developed costing methods using linked cancer registry and health care administrative data in adults in Canada [24–26]. The objective of this study was to use this published costing methodology to estimate total and net health care costs for 90 days prediagnosis (to capture tests and procedures before the definitive cancer diagnosis) and for 1 year postdiagnosis (when initial treatment is provided) in children and adolescents diagnosed with cancer in Ontario.

Methods

We conducted the analysis from the perspective of the public payer, the Ontario Ministry of Health and Long-Term Care. Our primary outcome measures were total and net direct medical costs borne by the public payer. The study was approved by the Research Ethics Boards of the University of Toronto and Sunnybrook Health Sciences Centre.

Data

We used health care administrative databases containing information on all Ontario residents, held at the Institute for Clinical Evaluative Sciences (ICES). We also used the Pediatric Oncology Group of Ontario Network Information System (POGONIS), which contains demographic, clinical, and treatment data from Ontario's five pediatric cancer centers, to which almost all Ontario children with cancer are referred. POGONIS has been shown to capture 96% of all patients diagnosed with cancer at ages 0 to 14 years [27]. There are no specialized adolescent or young adult cancer centers in Ontario, and POGONIS was shown to capture only 48% of adolescents diagnosed at ages 15 to 17 years [27]. Therefore, we used the population-based Ontario Cancer Registry, which includes records of 98% of all newly diagnosed cancers in Ontario, except for nonmelanoma skin cancer [28,29], to complete the identification of patients aged 15 years and older. Cancer Care Ontario provided additional data on radiation therapy and chemotherapy. All data sets were linked using unique encoded identifiers and analyzed at ICES.

Patients

We identified and analyzed two groups of patients with cancer diagnosed between January 1, 1995, and December 31, 2009: children, diagnosed at age 91 days (to have data for 90 days prediagnosis) to 14 years, and adolescents, diagnosed at age 15 to 19 years. Both cohorts received an initial diagnosis of 1 of the 12 groups of the International Classification of Childhood Cancer, Third Edition [30] (ICCC). Children were identified from POGONIS, and adolescents were identified from POGONIS and the Ontario Cancer Registry. When duplicate records were found for adolescents, we selected the record from the Ontario Cancer Registry over the POGONIS record for data related to diagnosis, because this resulted in most patients being selected from the same source. We excluded patients with identical dates of diagnosis and death, or with missing or invalid OHIP number, histology code, or sex. We followed patients for 1 year after diagnosis, or from diagnosis to death if they died within the year. We stratified patients into those who survived for less than a year and those who survived for a year or longer, because previous work in adults indicated that costs of care differ by postdiagnosis survival [26].

Patients were described in terms of cancer type, comorbidity, and demographic characteristics. We described comorbidity by categorizing all *International Classification of Diseases* (*Ninth Revision, Ninth Revision-Clinical Modification, and Tenth Revision*) diagnostic codes in hospital records and physicians' billing data in the year before diagnosis, or from birth to diagnosis for patients diagnosed when aged less than 1 year, into 1 of 29 aggregated diagnosis groups (ADGs), a unit in a population-patient case-mix adjustment system [31] that relates to health care costs in patients of all ages [32]. We classified the total number of ADGs assigned to each patient into the following groups: 0, 1 to 4, 5 to 9, and 10 or more. We used the Statistics Canada Postal Code Conversion file and data from the 2001 Canada Census to obtain neighborhood-level median household income (measured in quintiles) and rurality of residence (rural/urban). Communities with a population of less than 10,000 were defined as rural [33,34].

We classified children into the three most common diagnostic groups (leukemia, lymphoma, and central nervous system [CNS] tumors), and a fourth category for "other" cancers, for most of our analyses. In addition, we examined net costs in the 12 ICCC groups. We distinguished seven major diagnostic groups in adolescents: leukemia, lymphoma, CNS tumors, bone tumors and soft tissue sarcomas, germ cell tumors, thyroid carcinomas, and other cancers. In addition, we examined net costs in adolescents with acute lymphoblastic leukemia (ALL) [35], and those with other types of leukemia, as well as those in the other ICCC groups.

Controls

We used the Ontario Registered Persons Database, which contains information on all persons registered for OHIP coverage, to select controls without cancer. We estimated net cancer-related costs by subtracting the total health care costs of controls from the total health care costs of patients with cancer [25,36,37]. Potential controls were matched on birth year and month, and assigned an index date corresponding to the diagnosis date of the patient. We computed the propensity score (probability of being a patient with cancer) for each patient and potential control using logistic regression, with sex, rurality, and number of ADGs as predictors [38]. Greedy matching was used to select three controls