#### RESEARCH PAPER

# Comparing Childhood Cancer Care Costs in Two Canadian Provinces

Comparaison des coûts associés aux soins oncologiques chez les enfants dans deux provinces canadiennes



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[76] HEALTHCARE POLICY Vol.15 No.3, 2020

# Abstract

*Background*: Cancer in children presents unique issues for diagnosis, treatment and survivorship care. Phase-specific comparative cost estimates are important for informing healthcare planning.

*Objectives*: The aim of this paper is to compare direct medical costs of childhood cancer by phase of care in British Columbia (BC) and Ontario (ON).

*Methods*: For cancer patients diagnosed at <15 years of age and propensity-score-matched non-cancer controls, we applied standard costing methodology using population-based healthcare administrative data to estimate and compare phase-based costs by province. *Results*: Phase-specific cancer-attributable costs were 2%–39% higher for ON than for BC. Leukemia pre-diagnosis costs and annual lymphoma continuing care costs were >50% higher in ON. Phase-specific in-patient hospital costs (the major cost category) represented 63%–82% of ON costs, versus 43%–73% of BC costs. Phase-specific diagnostic tests and procedures accounted for 1.0%–3.4% of ON costs and 2.8%–13.0% of BC costs.

*Conclusions*: There are substantial cost differences between these two Canadian provinces, BC and ON, possibly identifying opportunities for healthcare planning improvement.

# Résumé

Contexte : Le cancer chez l'enfant présente des problèmes uniques en matière de diagnostic, de traitement et de survie. Il importe d'effectuer une comparaison portant sur l'estimation des coûts, selon des étapes précises, pour renseigner la planification des services de santé. Objectif : L'objectif de cet article est de comparer les coûts médicaux directs du cancer chez l'enfant, selon les étapes de soins, en Colombie-Britannique et en Ontario. Méthode : Pour les patients qui ont reçu un diagnostic de cancer avant l'âge de 15 ans et pour le groupe témoin de personnes non cancéreuses au score de propension similaire, nous avons employé une méthodologie standard pour le calcul des coûts au moyen des données administratives de santé afin d'estimer et de comparer, étape par étape, les coûts dans les provinces. Résultats : Les coûts attribuables au cancer pour les étapes à l'étude étaient de 2 à 39 % plus élevés en Ontario qu'en Colombie-Britannique. Les coûts pré-diagnostics associés à la leucémie et les coûts annuels pour un lymphome étaient >50 % plus élevés en Ontario. Les coûts des patients hospitalisés pour les étapes à l'étude (la principale catégorie de coûts) représentaient de 63 à 82 % des coûts en Ontario, contre 43 à 73 % en Colombie-Britannique. Les tests et procédures diagnostiques pour les étapes à l'étude comptaient pour 1,0 à 3,4 % des coûts en Ontario, contre 2,8 à 13,0 % en Colombie-Britannique.

*Conclusion* : Il y a d'importantes différences de coûts entre les deux provinces canadiennes, l'Ontario et la Colombie-Britannique, ce qui laisse possiblement place à une amélioration dans la planification des services de santé.

# Introduction

The cost of treating cancer in Canada is increasing (de Oliveira et al. 2018). Healthcare funders must deliver high-quality and high-value care within sustainable healthcare systems. Assessment and comparison of performance of different healthcare systems, using recognized indicators such as patient-level utilization and costs over a disease course (Carinci et al. 2015), can identify opportunities for care and system improvement (Tarricone 2006). Differences between systems in policy/practice factors (e.g., locus of care, resource distribution and staff mix) and exogenous factors (e.g., labour costs, patient mix, facility size and remoteness) can point to opportunities to improve resource allocation, system performance, accessibility and sustainability (CIHI 2017; Cunningham 2000; Lipscomb et al. 2013; Robinson 1993; Tarricone 2006). Adult cancer-related resource distribution and costs were found to vary between Ontario (ON) and British Columbia (BC; de Oliveira et al. 2017a). Cancer costs for children likely differ from those for adults (CCS 2017; Ellison et al. 2009) because children have distinct cancer types often requiring lengthy complex therapy (Lanzkowsky et al. 2016; Nathan et al. 2019) at pediatric cancer centres, which provide more comprehensive care than adult facilities (Brand et al. 2016). Also, overall survival is higher (CCS 2017; Chan and Raney 2005; Ellison et al. 2009) among children, and most survivors experience late and/or long-term effects of cancer and/or its treatment (Lorenzi et al. 2011; Oeffinger et al. 2006).

Few studies have measured direct medical costs of childhood cancer care in Western countries (de Oliveira et al. 2017b, 2017c; Luo et al. 2002; Mueller et al. 2017; Nathan et al. 2019; Price et al. 2012). This observational study estimates cancer-attributable healthcare costs for children with cancer by phase of care in the provinces of ON and BC, using administrative data, to identify potentially policy-relevant interprovincial differences.

# Methods

### Ethics approvals and data management

The study was approved by institutional review boards at Sunnybrook Health Sciences Centre and the University of Toronto, Toronto, ON, and at BC Cancer/University of British Columbia, Vancouver, BC. Data set access was approved by data stewards (ministry individuals authorized to approve research access to government data). Identifying information was removed; individual consent was not required. ON and BC data were analyzed separately using parallel procedures, recognizing differences between provincial data sets.

## Subjects

Eligible cases were residents of BC and ON with a first diagnosis of any cancer or tumour from January 1995 to June 2010 based on the International Classification of Childhood Cancer, Third Edition (Steliarova-Foucher et al. 2005), before the age of 15 years. These provinces represent 49.8% of the Canadian pediatric population and were the only Canadian provinces with multiple years of detailed comparable data. ON cases were identified from the Paediatric Oncology Group of Ontario Network Information System, a registry and clinical database from the five ON pediatric cancer centres, which treat 96% of ON children diagnosed at <15 years (Greenberg et al. 2003). BC cases were identified from the BC Cancer Registry, which ascertains all cases diagnosed among BC residents using multiple sources, including the BC Children's Hospital (BCCH) and BC cancer treatment centres. Over 90% of children with cancer are referred to the BCCH. Cases with identical dates of diagnosis and death, missing or invalid codes for histology or sex or invalid provincial medical insurance plan numbers were excluded. We grouped patients into the three most common childhood cancers (leukemia, lymphoma and central nervous system [CNS] tumours) and a fourth group consisting of all other cancers.

Control subjects without cancer were selected from the provincial health insurance plan registries. Potential controls were randomly matched to cases on sex, birth year and month and assigned index dates corresponding to the diagnosis dates of the cases. A propensity score (probability of having cancer) was computed for each case and potential control, with sex, neighbourhood-level rurality of residence (Statistics Canada 2013) and co-morbidity as predictors. Co-morbidity was calculated for the year before diagnosis/index date, or from birth to diagnosis/index date if less than a year, using number of Aggregated Diagnosis Groups, a morbidity grouping generated from diagnostic codes in in-patient and outpatient administrative records, in a case-mix adjustment system validated for patients of all ages (Reid et al. 2001; Starfield and Kinder 2011). "Greedy matching" was used to select three controls with the closest propensity scores within 0.1 standard deviation of each patient's propensity score (Austin 2011; Austin and Mamdani 2006). Selected controls lived at least as long as the patients to whom they were matched.

#### Costs and data sources

Direct medical costs were tracked from 60 days before diagnosis or index date or from date of birth (whichever was later) until death or December 31, 2010, the latest date for which all data sets were available in both provinces. Costs from all years were converted to 2012 Canadian dollars using the Consumer Price Index (Statistics Canada 2012).

Data sets included information on in-patient hospitalizations, same-day surgery/procedures, chemotherapy, radiotherapy, physician services, diagnostic tests, home and community care, complex continuing care and outpatient prescription drugs. Supplementary Table 1 describes the data and costing methods and can be found at https://www.longwoods.com/ content/26129.

Costs for hospital-based services were estimated by multiplying the resource intensity weight, a measure of resource utilization (CIHI 2016), of each visit by the cost per weighted case (CPWC), the cost of treating an average patient for a specific hospital (Wodchis et al. 2013), for the year of use (de Oliveira et al. 2017b, c). Because children have complex care needs, pediatric hospitals have higher CPWC values than general hospitals. Therefore, cost

estimates for in-patient hospitalizations, emergency department (ED) visits and same-day surgeries were generated using hospital-specific CPWC of pediatric hospitals (de Oliveira et al. 2017b, c) and the provincial average hospital CPWC for other hospitals.

# Statistical analysis

Based on clinical relevance and joinpoint analysis (Baker et al. 1991; de Oliveira et al. 2017a; Kim et al. 2000; Yabroff et al. 2008), we defined (1) pre-diagnosis phase, including diagnostic testing, as the 60 days prior to diagnosis (or from birth to diagnosis in patients diagnosed before 60 days of age); (2) initial phase, encompassing primary treatment, from diagnosis date to 360 days after diagnosis; (3) continuing phase of variable length, involving surveillance and follow-up care; and (4) terminal phase, up to 360 days before death, for those who died. The continuing phase included the time between the end of the initial phase and the start of the terminal phase or the end of the study observation period (December 31, 2010). For terminal phase determination, we looked forward to December 31, 2011, to see if patients died within one year after the end of the observation period. Phase-specific mean costs per patient, including patients who did not use the service and had zero cost, and 95% confidence intervals were estimated overall, by cancer type and resource, for cases and controls. Costs for initial, continuing and terminal phases were standardized to annual costs, based on a 360day year. Mean net (cancer-attributable) costs were estimated using a generalized estimating equation model (SAS Proc GENMOD), with case and control as binary clusters, to generate individual costs.

# Results

## Subjects

There were 1,503 cases (4,509 controls) in BC and 4,606 cases (13,818 controls) in ON available for analysis (Supplementary Table 2, available at https://www.longwoods.com/ content/26129). Approximately 92% of cancer patients (1,390BC; 4,261ON) were part of the initial phase, 85% (1,293BC; 3,880ON) spent time in the continuing phase, and only 18%–19% (273BC; 893ON) entered the terminal phase. Sociodemographic characteristics and co-morbidity were similar between BC and ON cases.

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| Cancer                           |              |               |           |            |           |  |
|----------------------------------|--------------|---------------|-----------|------------|-----------|--|
| diagnosis                        | N (% )       | Pre-diagnosis | Initial   | Continuing | Final     |  |
| British Columbia                 |              |               |           |            |           |  |
| Leukemia*                        | 494 (32.9)   | \$2,879       | \$107,129 | \$14,436   | \$523,870 |  |
| Lymphoma*                        | 151 (10.0)   | \$5,264       | \$81,650  | \$4,865    | \$643,418 |  |
| Central nervous<br>system (CNS)* | 345 (23.0)   | \$7,453       | \$84,372  | \$10,193   | \$231,412 |  |
| Other§                           | 513 (34.1)   | \$5,049       | \$105,455 | \$16,200   | \$208,000 |  |
| Total                            | 1,503 (100)  | \$4,909       | \$99,087  | \$13,133   | \$310,798 |  |
| Ontario                          |              |               |           |            |           |  |
| Leukemia*                        | 1,636 (35.5) | \$4,898       | \$156,225 | \$21,418   | \$432,010 |  |
| Lymphoma*                        | 459 (10.0)   | \$5,587       | \$96,380  | \$7,462    | \$373,492 |  |
| CNS*                             | 980 (21.3)   | \$7,910       | \$115,056 | \$10,878   | \$282,738 |  |
| Other§                           | 1,531 (33.2) | \$7,993       | \$144,911 | \$14,970   | \$249,384 |  |
| Total                            | 4,606 (100)  | \$6,637       | \$138,161 | \$15,756   | \$316,303 |  |

**TABLE 1.** Phase-specific mean net costs by diagnosis\* (Canadian \$2012) for childhood cancer cases(1995–2010) in British Columbia and Ontario

\* International Classification of Childhood Cancer (ICCC), Third Edition (Steliarova-Foucher et al. 2005)

§ All other nine ICCC groups

#### Costs

For all cancers combined, mean net costs in ON were higher than those in BC, across all phases of care (Table 1). The excess ranged from 1.8% (for the terminal phase) to 39.4% (for the initial phase). In both provinces, the highest mean net costs were in the terminal phase, followed by the initial phase. Pre-diagnosis (60-day) costs represented between 1.6%BC and 2.0%ON of terminal care (360-day) costs; continuing care costs totalled between 4.2%BC and 5.0%ON of terminal care costs.

| TABLE 2. | Phase-specific mean | net costs | by resource | (Canadian | \$2012) f | or British | Columbia | and |
|----------|---------------------|-----------|-------------|-----------|-----------|------------|----------|-----|
| Ontario  |                     |           |             |           |           |            |          |     |

| Resource                        | Pre-diagnosis | Initial   | Continuing | Final     |  |  |  |
|---------------------------------|---------------|-----------|------------|-----------|--|--|--|
| British Columbia                |               |           |            |           |  |  |  |
| In-patient hospitalization      | \$2,146       | \$70,371  | \$7,016    | \$225,939 |  |  |  |
| Physician services              | \$1,832       | \$8,610   | \$847      | \$46,826  |  |  |  |
| Diagnostic tests and procedures | \$626         | \$4,784   | \$864      | \$8,842   |  |  |  |
| Chemotherapy                    | N/A           | \$7,476   | \$1,105    | \$8,630   |  |  |  |
| Radiotherapy                    | N/A           | \$2,677   | \$285      | \$6,236   |  |  |  |
| Total*                          | \$4,909       | \$99,087  | \$13,133   | \$310,798 |  |  |  |
| Ontario                         |               |           |            |           |  |  |  |
| In-patient hospitalization      | \$4,735       | \$106,660 | \$9,943    | \$260,872 |  |  |  |
| Physician services              | \$586         | \$7,201   | \$1,170    | \$20,463  |  |  |  |
| Diagnostic tests and procedures | \$273         | \$1,581   | \$536      | \$3,120   |  |  |  |
| Chemotherapy                    | N/A           | \$13,240  | \$2,211    | \$10,519  |  |  |  |
| Radiotherapy                    | N/A           | \$2,315   | \$132      | \$7,194   |  |  |  |
| Total*                          | \$6,177       | \$138,161 | \$1,556    | \$316,303 |  |  |  |

\*Includes resources listed above plus emergency department visits, same-day surgery, outpatient prescription drugs, home/community care and complex continuing care.

The ranking of the magnitude of diagnosis-specific costs across the phases was similar in both provinces (Table 1). Pre-diagnosis costs were highest for patients with CNS tumours and lowest for those with leukemia. The initial phase costs were highest for leukemia patients and lowest for patients with lymphomas. For the continuing phase, the highest costs were for patients with leukemia in ON and "other" cancers in BC, but in both provinces, patients with lymphoma had the lowest costs. For the terminal phase, the highest costs were for the leukemia patients in ON and the lymphoma patients in BC, and the lowest costs were for patients with "other" cancers in both provinces.

Higher diagnosis-specific costs were seen in ON than in BC for most phases of care. For leukemia patients, pre-diagnosis costs were 59% higher, initial treatment costs were 46% higher and annual continuing care costs were 48% higher. Terminal phase costs for leukemia patients were 18% lower in ON. For lymphoma patients, the provincial differences were not as large or consistent (ON 3% lower than BC in pre-diagnosis; 18% higher than BC for initial treatment; 53% higher than BC for continuing care; and 42% lower than BC for terminal care). Costs in ON were slightly higher than those in BC for CNS tumour patients; specifically, pre-diagnosis costs were 2% higher, initial treatment costs were 36% higher, continuing care costs were 7% higher and terminal phase costs were 22% higher. Among patients with "other" cancers, pre-diagnosis and initial treatment costs were 44% and 37% higher, respectively, in ON. Continuing care costs were 7% lower in ON, and terminal phase costs were 20% higher in ON.

The distribution of costs by resource varied between the provinces in all phases (Table 2). In-patient costs comprised a larger proportion of total costs in ON: pre-diagnosis (77%ON vs. 43%BC), initial (77%ON vs. 71%BC), continuing (63%ON vs. 52%BC) and terminal (82%ON vs. 73%BC). In contrast, physician services accounted for 7.4%-13.0% of ON costs and 6.4%–37.0% of BC costs, and diagnostic tests and procedures accounted for 1.0%–3.4% of ON costs and 2.8%–13.0% of BC costs. Notably, in the pre-diagnosis phase, 9.5% of ON costs, but 37% of BC costs, were for physician services, but in the continuing phase, 74% of ON costs were for physician services (vs. 6.4% in BC). The mean and proportion of day-surgery costs in continuing care (2.4%ON vs. 17.0%BC) were much lower in ON. The proportion of costs related to chemotherapy was higher in ON than in BC for the initial (9.6%ON vs. 7.5%BC), continuing (14.0%ON vs. 8.4%BC) and terminal (3.3%ON vs. 2.8%BC) phases, but the proportion attributed to radiotherapy was lower in ON than in BC for the initial (1.7%ON vs. 2.7%BC) and continuing (0.8%ON vs. 1.9%BC) phases. Net costs (total costs for cases minus costs for controls) were only slightly lower than total costs for all phases, in BC and ON, indicating that most costs for the cancer cases were cancer-related (Table 3).

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| Phase         | British Columbia |               | Ontario         |               |  |
|---------------|------------------|---------------|-----------------|---------------|--|
|               | Mean Total Cost  | Mean Net Cost | Mean Total Cost | Mean Net Cost |  |
| Pre-diagnosis | \$4,997          | \$4,909       | \$6,637         | \$6,177       |  |
| Initial       | \$99,648         | \$99,087      | \$139,453       | \$138,161     |  |
| Continuing    | \$13,607         | \$13,133      | \$16,786        | \$15,756      |  |
| Terminal      | \$311,398        | \$310,798     | \$319,883       | \$316,303     |  |

**TABLE 3.** Phase-specific mean total and net costs (Canadian \$2012), for childhood cancer cases inBritish Columbia and Ontario

Net costs = Total costs for cases minus costs for controls

### Discussion

This study compared cancer-attributable (net) childhood cancer medical costs in two Canadian provinces with publicly funded, comprehensive healthcare systems covering virtually all residents. We used the same criteria to select patients and used similar population-based registries and healthcare administrative data sets over the same period. Overall mean phase-specific costs were higher for ON than for BC, mainly because of higher in-patient hospital costs (the major cost category) in ON.

In both provinces, costs were highest in the terminal and initial care phases, reflecting cost patterns observed in adult cancer patients in Canada (de Oliveira et al. 2017a) and the US (Brown et al. 2002). However, costs were much higher for children because of the duration and intensity of childhood cancer treatments (Lanzkowsky et al. 2016). Childhood cancer-related diagnostic procedures, treatment protocols and follow-up care guidelines are highly standardized in North America but vary by diagnosis (Lanzkowsky et al. 2016). Consequently, we observed cost differences by diagnosis within provinces but similar diagnosis-specific costs between provinces.

The large proportion of hospital-based costs in our study is consistent with that reported in previous studies (Luo et al. 2002; Mueller et al. 2017; Rosenman et al. 2005). Most of the hospital visits in both provinces were at tertiary/quaternary centres for oncology care for individuals aged 18 years or younger. These centres would see patients with similar distributions of severity of disease, deliver similar care and have higher CPWC than other hospitals. ON's The Hospital for Sick Children has an estimated CPWC over twice as high as most other ON hospitals (KPMG 2012), and the estimated CPWC for BCCH is almost 60% higher than that for other BC hospitals. Furthermore, higher proportions of ON than BC patients were hospitalized in the initial (4.5% higher) and terminal (7.9% higher) phases, which would increase costs substantially. Despite similar proportions of users of physician services, diagnostic tests and procedures between provinces (data not shown), costs in BC were >50% lower for the initial and terminal phases, which could be related to fewer or less costly encounters per user. Differences in standard practice may partly explain these differences. There are more restrictive criteria for requesting clinical investigations, and more investigations undertaken as outpatients, in BC (personal communication with BC pediatric oncologist co-author [PCR]). There may also be inter-centre variations in patterns of care in

ON. These differences may contribute to lower hospital costs and higher physician services and diagnostic services costs in BC compared to in ON.

This study used geographically defined population-based administrative data sets documenting "medically necessary care" (Madore 2005). The systems in BC and ON are more similar to each other than to the system in Quebec, another large Canadian province, whereas the provinces with smaller populations have fewer well-developed data resources. Data sets included costs for most of the resources covered by provincial governments. This approach provides more accurate and comparable estimates of direct medical care costs than indirect data sources and non-representative populations. The "phase of care" approach is relevant to clinical care and patient experience.

Documentation of cost and utilization patterns of healthcare is fundamental to identifying opportunities to improve practice and policy. The next step is to explore the policy/ practice-related and exogenous components of inter-provincial cost variations, including delivery models, resource issues, patient mix and socio-economic and geographic factors. A recent ON report on comparative hospital costs (CIHI 2017) concluded that most cost differences could be explained by exogenous factors or hospital management decisions, related more to system and delivery model factors than to institutional factors (CIHI 2017). More research has yet to be done to explore the results of the BC–ON adult study (de Oliveira et al. 2017a); therefore, no policy or practice changes have followed to date.

Some differences in costs may be owing to reporting artifacts. Some histology and diagnostic tests, counted in the ON claims data, were not recorded in the BC claims database because these are covered by institutional budgets. Some data sources were not comparable between provinces (e.g., ED visits, home care and outpatient prescription drugs). The proportion of patients with ED visits was similar between provinces in the pre-diagnosis and initial phases, whereas the proportion of users of home care varied dramatically between provinces, suggesting that home-care data sources might be measuring very different types of care or patient subgroups (data not shown). Although costs for in-patient hospitalization and physician services were estimated from identical or very similar databases in each province, some cost variation can be attributed to differences in the scope of services covered and recorded, as well as to difficulties in harmonizing data, missing data, changes in data capture over time and variable levels of data to estimate costs.

Time from data set availability to analysis completion resulted in an interval to reporting. However, five-year survival estimates for childhood cancer have been 82%–83% from 1999 onward (CCS 2008; CCS 2017), and the protocols generating these survival rates have not changed appreciably in recent years. We followed patients for up to 15 years after diagnosis, thus capturing costs of late effects of treatment within that time, but studies that tracked survivors of childhood cancer for up to 40 years following the diagnosis found that relative risks for late effects increased as patients aged (Geenen et al. 2007; Hudson et al. 2013; Lorenzi et al. 2011; Oeffinger et al. 2006). Therefore, net continuing care costs may change over time (Luo et al. 2002). Also, caregivers and survivors have many additional ongoing costs, including out-of-pocket costs relating to loss of work days, treatment-related travel and family supports (Warner et al. 2015).

In conclusion, this large population-based study compared costs of childhood cancer care in two jurisdictions with similar healthcare systems and observed cost variation that was potentially modifiable, once determinants of variation are understood, in particular relating to resource allocation and system efficiencies. This type of analysis should provide useful guidance for other Canadian provinces, all of which operate under the Canada Health Act, and other within-country childhood cancer cost evaluations. The significant challenges in conducting cross-national comparative cost studies, in particular because of variation in data measurement, limit our current ability to produce valid comparisons among countries (Lipscomb et al. 2013). Although childhood cancer is rare (<1% all cancers; CCS 2008, 2017), survival in Canada and many Western nations is high (>80% overall) (CCS 2008; Ward et al. 2014) largely because of successful treatment, with many person years of life saved (CCS 2017; Chan and Raney 2005; Ellison et al. 2009). Therefore, strategies and interventions to improve care are likely to be cost effective; however, assessment of effect on patient survival and other outcomes is required to examine this question. Future work should assess costs for additional years in continuing care and update costs to account for newer treatments, to identify additional opportunities for improved system performance. Overall, comparative cost studies highlight differences in healthcare in different jurisdictions, and comparative results can support healthcare research and policy and care improvement.

#### Conflict of Interest Statement:

The authors have no conflicts of interest or relevant financial disclosures.

#### Acknowledgements

All inferences, opinions and conclusions drawn in this publication are those of the authors and do not reflect the opinions or policies of the British Columbia Data Steward(s) (British Columbia Ministry of Health 2011a, b, c, d, e, f).

In Ontario, this work was supported by the Institute for Clinical Evaluative Sciences, which is funded by an annual grant from the Ontario Ministry of Health and Long-Term Care (MOHLTC). The opinions, results and conclusions reported in this paper are those of the authors and are independent from the funding sources. No endorsement by Institute for Clinical Evaluative Sciences or the Ontario MOHLTC is intended or should be inferred.

Parts of this material are based on data and information compiled and provided by the Canadian Institute for Health Information (CIHI). However, the analyses, conclusions, opinions and statements expressed herein are those of the authors and not necessarily of CIHI.

Parts of this material are based on data and information provided by Cancer Care Ontario (CCO). The opinions, results, views and conclusions reported in this paper are those of the authors and do not necessarily reflect those of CCO. No endorsement by CCO is intended or should be inferred. Correspondence may be directed to: Mary L. McBride, MSc, Cancer Control Research, BC Cancer, 2-106, 675 West 10th Avenue, Vancouver, BC, V5Z 1L3. She can be reached by phone at 604-802-6658. Her e-mail address is mmcbride@bccrc.ca.

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