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Predictors of attendance at specialized survivor clinics in a population-based cohort of adult survivors of childhood cancer

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Abstract
Purpose The purpose of the present study is to determine predictors of attendance at a network of publicly funded specialized survivor clinics by a population-based cohort of adult survivors of childhood cancer.
Methods We conducted a retrospective study linking data on eligible patients identified in a provincial pediatric cancer registry with health administrative databases to determine attendance at five specialized survivor clinics in the Canadian province of Ontario between 1999 and 2012. Eligible survivors were treated for cancer at ≤18 years between 1986 and 2005, had survived ≥5 years from their most recent pediatric cancer event, and contributed ≥1 year of follow-up after age 18 years. We assessed the impact of cancer type, treatment intensity, cumulative chemotherapy doses, radiation, socioeconomic status, distance to nearest clinic, and care from a primary care physician (PCP) on attendance using recurrent event multivariable regression.
Results Of 7482 children and adolescents treated for cancer over the study period, 3972 were eligible for study inclusion, of which 3912 successfully linked to administrative health data. After a median of 7.8 years (range 0.2–14.0) of follow-up, 1695/3912 (43.3 %) had attended at least one adult survivor clinic visit. Significantly increased rates of attendance were associated with female gender, higher treatment intensity, radiation, higher alkylating agent exposure, higher socioeconomic status, and an annual exam by a PCP. Distance significantly impacted attendance with survivors living >50 km away less likely to attend than those living within 10 km (relative rate 0.77, $p = 0.003$).
Conclusion Despite free access to survivor clinics, the majority of adult survivors of childhood cancer do not attend.
Implications for Cancer Survivors Alternate models of care need to be developed and assessed, particularly for survivors living far from a specialized clinic and those at lower risk of developing late effects.

Keywords Survivor clinics · Models of care · Childhood cancer survivors · Administrative health databases · Cancer registries

Introduction

Significant improvements in the probability of survival in children with cancer have resulted in a burgeoning population of long-term survivors. Many of the almost 400,000 childhood cancer survivors alive in the USA [1] are at risk for chronic morbidity and premature mortality as a consequence of their cancer treatment; 80 % of survivors will develop one or more severe or life-threatening chronic health conditions by the age of 45 years [2]. Consequently, it has been broadly accepted that survivors require life-long risk-adapted health care aimed at health promotion and periodic surveillance in order to prevent or mitigate the development of late effects [3, 4].

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Several models have been proposed for the care of survivors [5]. Among these, dedicated care at a specialized survivor clinic (usually in a cancer center) or shared care between a survivor clinic and a primary care physician (PCP) [6] has been advocated, particularly for survivors at higher risk for developing late effects. In Ontario, Canada's most populous province, the provincial government has funded a network of childhood cancer survivor clinics since 1999. Seven specialized clinics operate in the five cities that house the province's pediatric cancer centers. In three cities, a single clinic provides care to both children and adults who have survived childhood cancer. In two cities, survivors are transferred at age 18 years from a clinic situated in a pediatric hospital to a clinic situated in a separate adult cancer center. These clinics are accessible to survivors at no cost through Ontario's publically funded health care system. Although almost every eligible survivor attends such a clinic as a child or adolescent, many do not continue to attend these clinics once they reach adulthood. In the present study, we assessed the patterns and predictors of specialized survivor clinic attendance by adult survivors of childhood cancer.

Methods

Study population

After obtaining Research Ethics Board approval, we conducted a retrospective study of a population-based cohort of adult survivors of childhood cancer in Ontario. Eligible participants were identified in the Pediatric Oncology Group of Ontario Networked Information System (POGONIS), an active registry of all children and adolescents treated for cancer at any of the province's five pediatric cancer centers. Patients were eligible for inclusion if they were an Ontario resident diagnosed with cancer prior to age 18 years between January 1st, 1986, and December 31, 2005, had survived ≥5 years from their most recent pediatric cancer event, had at least 1 year of follow-up as an adult (≥18 years), and had contributed follow-up time after January 1, 1999 (when the clinics were launched). The most recent pediatric cancer event was defined as the latest of the initial cancer diagnosis or any relapse or subsequent malignant neoplasm [SMN] that occurred prior to age 18 years. This allowed survivors of relapse or SMN during childhood to be included in the cohort. Subsequently, each survivor's index date was defined as the latest of the date at which they reached 5 years from the most recent pediatric event and their 18th birthday. Patients were followed from their index date until relapse, SMN, death, or end of study on December 31, 2012 (whichever occurred first).

Demographic, diagnostic, and treatment data in POGONIS were linked deterministically (using unique provincial health card numbers which were then encoded) to demographic and health administrative databases (Registered Persons Database [RPDB], Canadian Institution for Health Information Discharge Abstract Database, Ontario Health Insurance Plan [OHIP], and the National Ambulatory Care Reporting System) housed at the Institute for Clinical Evaluative Sciences. These databases capture information on ambulatory and hospital-based medical care received in the province, vital statistics, and current patient addresses which are updated at the time of receipt of any hospital-based care.

Outcome definition

We used the OHIP physician billings database to capture outpatient medical visits. We identified visits to survivor clinics by compiling a list of the unique OHIP billing numbers of all physicians who provided care at a survivor clinic during the study period. Since many of these physicians also provide acute oncology or radiation oncology care, we created an algorithm to differentiate visits for cancer therapy (e.g., in the case of relapse or SMN as an adult) from survivor clinic visits. Survivors who had evidence of an administrative code for a hospitalization or outpatient visit related to receipt of chemotherapy, radiation or palliative care or who had had greater than four visits to a regional cancer center within a 60-day period were designated as having had a relapse or SMN and not classified as having had a survivor clinic visit if seen by one of the designated physicians within that time period.

Covariate definitions

Cancers were classified into eight groups: acute lymphoblastic leukemia (ALL), acute myeloid leukemia (AML) and other leukemias, lymphoma, brain tumors, neuroblastoma, renal tumors, bone and soft tissue sarcomas, and other tumors. The overall intensity of cancer therapy was classified using the Intensity of Treatment Rating Scale version 3 (ITR-3) [7] on a four-level scale with 4 representing the most intensive therapy. Cumulative alkylating agent doses were converted to cyclophosphamide-equivalent doses (CED) [8] and divided into four groups. Similarly, anthracycline doses were expressed as doxorubicin-equivalent doses [9] and divided into three groups. Radiation therapy was classified as none, brain only, chest only, brain and chest, or other. We created a binary variable to describe patients who had suffered a second event (relapse, SMN) prior to age 18 years. We also created a binary variable to distinguish between patients diagnosed before and after the launch of the survivor clinics in 1999. We examined socioeconomic status (SES) [10] and distance to a specialized clinic [11], both variables that have been demonstrated to impact access to specialized health care services. The postal code corresponding to each survivor's primary residence was updated annually over the follow-up period. SES was defined using the Ontario Marginalization Index

which provides neighborhood-level measures of deprivation and marginalization based on the 2006 Canadian census measures of residential stability, material deprivation, ethnic concentration, and dependency [12]. This variable was classified into quintiles, with survivors living in neighborhoods in the first quintile considered to be the least deprived. We calculated the straight-line distance from current place of residence to the nearest survivor clinic. We examined the impact of the receipt of care from a PCP by determining whether each survivor had visited a PCP for an annual assessment in each 1-year period. Finally, we classified the model of survivor clinic at each subject's treating institution (combined pediatric/adult survivor clinic vs. separate pediatric and adult clinics).

Statistical analysis

We calculated proportions for categorical variables and medians for continuous variables. To investigate factors associated with the rate of clinic visits, we implemented an Andersen-Gill recurrent event multivariable regression model [13–15]. This model can be viewed as an extension of the Cox model that allows for repeated occurrences of a single type of event over time. Event times for each survivor were defined as the times from their index date to the dates of each clinic visit. A counting process data structure was created to implement the recurrent event model [15, 16]. Since the index date for some individuals was prior to the launch of the survivor clinics, discontinuous risk intervals were used to exclude the time from index to January 1, 1999, from the analysis [16]. A robust variance estimation approach was used to handle multiple visits by each survivor, and the Breslow method [15] was implemented to accommodate any ties in the clinic visit times across survivors. The p values for the regression estimates were obtained based on a Wald test using robust sandwich variance estimates [15].

The recurrent event regression model adjusted for all covariates listed above was conducted under a complete case analysis. Neighborhood SES, distance to the closest clinic, and annual physical exam were treated as time-varying covariates. All other covariates were treated as baseline (time-fixed) measures.

To illustrate the relationship between distance and clinic visits, we used a nonparametric estimation approach to compute the mean cumulative function [17]. We plotted the mean cumulative number of clinic visits over time for each category of distance to closest survivor clinic. Variables with p values less than 0.05 were considered to be significant. All analyses were completed using SAS 9.2 (Cary, NC).

Results

Of the 7842 children and adolescents diagnosed with cancer and treated at one of the province's pediatric cancer centers between 1986 and 2005, 3972 met the criteria for study inclusion and 3912 (98.5 %) were successfully linked to their administrative health data (Fig. 1). Their demographic, disease, and treatment characteristics are described in Table 1.

Survivors were followed for a median of 7.8 years (range 0.2–14.0) after their index date and contributed 32,029 person-years of follow-up. Only 1695/3912 (43.3 %) attended at least one survivor clinic visit as an adult. Among attendees, the median number of visits over the study period was 3 (range 1–30).

Table 2 displays the recurrent event multivariate regression model for clinic attendance. Statistically significant increased rates of attendance were associated with female gender; higher treatment intensity; radiation to the brain, chest, or other sites (compared to no radiation); higher alkylating agent exposure; lower SES; and an annual exam by a PCP. Decreased attendance was associated with a diagnosis of a brain tumor or other cancer (compared to ALL), diagnosis prior to the launch of the survivor clinics in 1999, treatment at a center with a combined pediatric/adult survivor program, and increasing distance from a survivor clinic. Survivors who lived >50 km from a clinic were significantly less likely to attend (Fig. 2). The mean straight-line distance to the nearest clinic was 84 km (standard deviation [SD] 294) among survivors who attended at least one clinic compared to 120 km (SD 384) among those that did not attend any clinics.

Discussion

In this population-based cohort of childhood cancer survivors treated over a 20-year period, less than half attended a specialized survivor clinic as an adult despite access to these services at no cost as a function of Canada's universal health care system. A survey of US survivors reported that with the introduction of the Patient Protection and Affordable Care Act, almost 90 % of respondents have health insurance [18]. Thus, lack of insurance should not be a barrier to clinic attendance for many survivors, although co-pays for medical visits and surveillance tests may be a barrier to low-income patients. Further, indirect costs (e.g., time off work, travel, etc.) may impact attendance, particularly since specialized survivor clinics are usually located in large urban centers. In the present study, distance from the closest clinic significantly impacted attendance—48 % of survivors living within 50 km of a clinic had at least one visit compared with only 35 % of those who lived more than 50 km away.

Several experts have advocated a tiered model of survivor care based on the intensity of cancer therapy and the consequent risk for late effects [4, 19, 20]. McCabe proposed a risk-stratified shared care model in which the survivor clinic provides direct care to the highest-risk survivors (e.g., those who have received high doses of alkylating or anthracycline

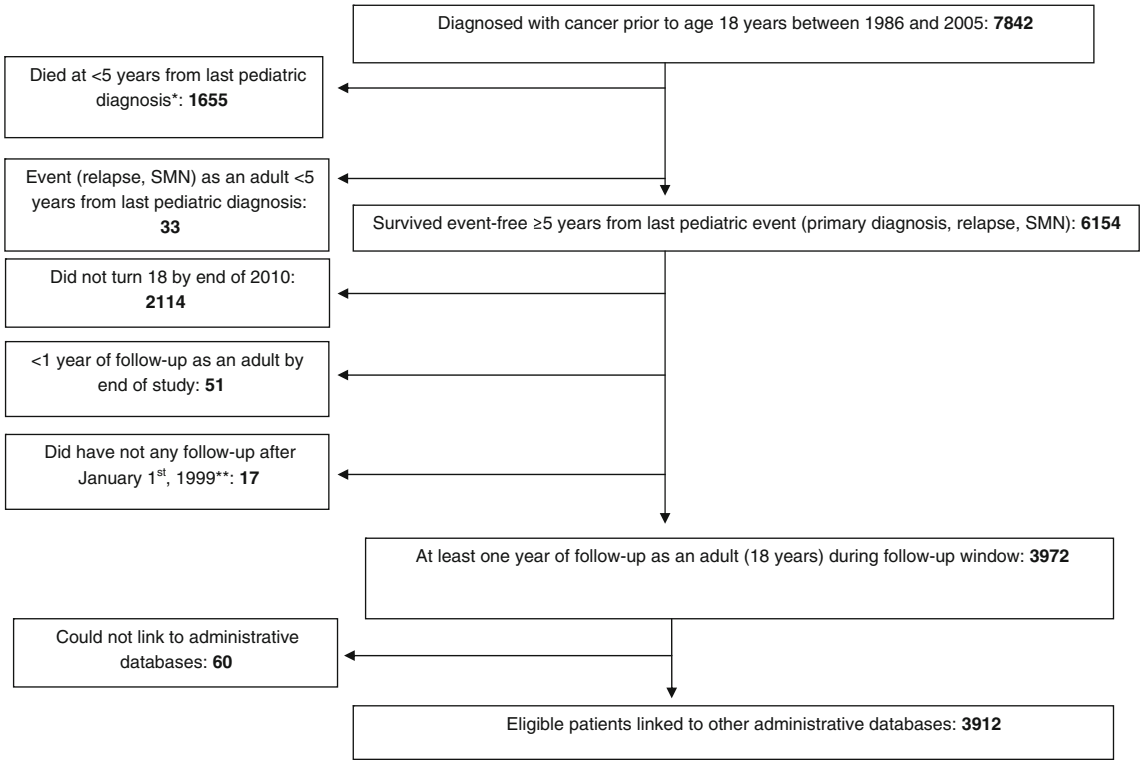

Fig. 1 Flow diagram of derivation of study cohort

agents, high-dose radiation, and allogeneic stem cell transplant and those who have persistent multi-organ toxicities of therapy) [20]. A PCP assumes primary responsibility for moderate- and low-risk patients, with the survivor program playing a supportive role. Although patients in the present study treated with more intensive therapy or with modalities known to increase the risk for late effects were more likely to attend a survivor clinic, only 41 % of survivors in the most intensively treated quartile had even a single clinic visit. Of particular concern was the observation that only 26 % of survivors of CNS tumors attended even a single appointment. Many of these survivors are at elevated risk for late effects (neurocognitive, endocrine, SMN, among others) as a consequence of cranial radiation or surgery, and some are unable to live or function independently as adults. Our findings mirror those observed in a recent publication describing specialized clinic attendance among survivors treated at single center in the USA [21]—in that cohort, only 7.2 % of CNS tumor survivors attended a specialized clinic, a proportion significantly lower that observed in other diagnostic groups. The reasons for this poor compliance with follow-up cannot be identified in the present analysis but require further investigation given the vulnerability of this cohort of survivors.

It is plausible that some survivors continue to receive care in the cancer center in an acute care oncology clinic (e.g., a stem cell transplant or brain tumor clinic), but these clinics frequently focus on surveillance for recurrence of the original cancer rather than the risks for late effects. Most survivors receive care only from a PCP. Unfortunately, surveys of North American family physicians [22] and US general internists [23] revealed that although approximately 85 % of PCPs are willing to care for childhood cancer survivors in collaboration with a cancer center, 63–77 % of respondents express discomfort with caring for this population. Only 1 % of family physicians and 6 % of general internists are willing to care for survivors independently. Knowledge regarding recommended surveillance is quite limited, with only 2 % of family physicians and 5 % of general internists being able to identify guideline-recommended cardiac, breast cancer, and thyroid function surveillance in a hypothetical survivor of childhood Hodgkin lymphoma. Survivors at increased risk for therapy-related cardiac dysfunction or breast cancer have been demonstrated to be less likely to receive guideline-recommended mammograms or echocardiograms if they are followed outside of a cancer center [24].

Data on the availability of specialized programs for adult survivors of childhood cancer in the USA is limited. A 2007 survey of 179 centers affiliated with the Children's Oncology Group revealed that only half offered care to cancer survivors in a specialized program during their pediatric years. Forty-four percent of programs retained their survivors as adults without transition to a PCP. Similar to the survivors in our

Table 1 Baseline characteristics (at the index date) of survivors who did and did not attend at least one survivor clinic over the follow-up period

	Attended clinic ($n=1695$)		Did not attend clinic ($n=2217$)		
	N	Row %	*N*	Row %	*p* value
Gender					
Female	794	44.4	993	55.6	0.20
Male	901	42.4	1224	57.6	
Age at diagnosis					
0–4	478	43.6	618	56.4	0.03
5–9	435	45.8	515	54.2	
10+	782	41.9	1084	58.1	
Socioeconomic status					
1 low deprivation	496	48.3	530	51.7	<0.0001
2	383	45.2	464	54.8	
3	312	42.9	415	57.1	
4	262	43.3	343	56.7	
5 high deprivation	196	35.5	356	64.5	
Unknown	46	29.7	109	70.3	
Primary diagnosis					
ALL	524	58.7	368	41.3	<0.0001
AML/other leukemias	76	44.4	95	55.6	
Lymphoma	389	50.5	382	49.5	
CNS	224	26.4	623	73.6	
Neuroblastoma	42	28.2	107	71.8	
Renal tumors	107	54.3	90	45.7	
Bone + STS	121	29.2	294	70.8	
Other	212	45.1	258	54.9	
Diagnostic period					
1986–1990	449	38.5	717	61.5	0.001
1991–1995	575	45.2	696	54.8	
1996–2000	407	46.1	476	53.9	
2001–2005	264	44.6	328	55.4	
Diagnosis prior to 1999					
No	1463	45.0	1791	55.0	0.0001
Yes	232	35.3	426	64.7	
Treatment intensity (ITR-3)					
1	124	24.2	389	75.8	$p<0.0001$
2	788	45.5	943	54.5	
3	575	59.6	389	40.4	
4	184	40.6	269	59.4	
Unknown	24	9.6	227	90.4	
Cyclophosphamide equivalent dose (mg/m^2)					
0	775	33.1	1563	66.9	0.0001
1–3999	386	57.2	289	42.8	
4000–7999	219	59.7	148	40.3	
8000+	315	59.2	217	40.8	
Doxorubicin equivalent dose (mg/m^2)					
0	637	30.1	1479	69.9	0.0001
1–249	648	59.2	446	40.8	
250+	410	58.4	292	41.6	
Radiation					
None	889	35.1	1641	64.9	0.0001
Brain only	324	58.1	234	41.9	
Brain/chest	99	47.4	110	52.6	
Chest only	90	68.7	41	31.3	
Other	293	60.5	191	39.5	
SMN or relapse before index date					
No	1562	43.7	2013	56.3	0.13
Yes	133	39.5	204	60.5	
Survivor clinic model					
Combined pediatric/adult program	474	44.7	587	55.3	0.30
Separate pediatric/adult programs	1221	42.8	1630	57.2	
Distance from survivor clinic					
< 10 KM	365	44.8	449	55.2	<0.0001
10–24 KM	417	49.5	425	50.5	
25–49 KM	434	48.2	467	51.8	
50–99 KM	287	41.9	398	58.1	
≥100 KM	192	28.7	478	71.3	
Complete history/physical examination by a PCP in the year preceding diagnosis					
Yes	328	19.4	408	18.4	0.45
No	1367	80.6	1809	81.6	

study, these adults are at risk for attrition from follow-up. Consequently, the barriers to providing appropriate care to adult survivors of childhood cancer appear to be threefold: (1) there is a paucity of specialized survivor clinics [25–27]; (2) even when such clinics exist, many survivors do not attend; and (3) most PCPs lack the comfort and knowledge to provide care to survivors, particularly in the absence of cancer center support [22, 23]. Efforts to improve PCP knowledge and comfort have included the creation of electronic survivor care plans that can be accessed by survivors and their PCP [28] and the development of a systematic approach to alternating visits between survivor clinics and PCPs [6].

Consistent with findings from other studies of access and use of health services [10], survivors with lower SES were less

Table 2 Recurrent event multivariable regression model examining the predictors of survivor clinic attendance

	Relative rates	95 % Confidence interval	*p* value
Gender			
Male (referent)			
Female	1.18	1.07–1.31	0.001
Age at diagnosis (years)	1.00	0.99–1.01	0.88
Socioeconomic status			
1	1.27	1.06–1.53	0.008
2	1.05	0.88–1.26	0.57
3	1.07	0.89–1.29	0.45
4	1.10	0.91–1.33	0.35
5 (referent)			
Primary diagnosis			
ALL (referent)			
AML + other leukemias	0.85	0.64–1.13	0.25
Lymphoma	1.11	0.93–1.32	0.24
Brain	0.63	0.50–0.77	<0.001
Neuroblastoma	0.76	0.48–1.21	0.25
Renal tumors	1.24	0.96–1.60	0.11
Bone + STS	0.77	0.59–1.02	0.06
Other	0.67	0.54–0.84	<0.001
Diagnosis prior to 1999			
No (referent)			
Yes	0.74	0.63–0.86	<0.001
Treatment intensity			
1 (referent)			
2	1.68	1.26–2.25	<0.001
3	2.21	1.65–2.96	<0.001
4	1.69	1.10–2.58	0.02
Cyclophosphamide equivalent dose (mg/m^2)			
0 (referent)			
1–3999	1.08	0.92–1.27	0.33
4000–7999	1.29	1.08–1.54	0.005
8000+	1.47	1.25–1.73	<0.001
Doxorubicin equivalent dose (mg/m^2)			
0 (referent)			
1–249	1.04	0.88–1.23	0.65
250+	1.08	0.91–1.28	0.35
Radiation			
None (referent)			
Brain only	1.44	1.23–1.67	<0.001
Brain/chest	1.22	0.95–1.56	0.12
Chest only	1.59	1.28–1.97	<0.001
Other sites	1.31	1.13–1.53	<0.001
SMN or relapse before index date			
No (referent)			
Yes	0.91	0.68–1.21	0.51
Survivor clinic model			
Separate pediatric/adult program (referent)			
Combined pediatric/adult programs	0.85	0.76–0.96	0.008
Distance from survivor clinic			
<10 KM (referent)			
10–24 KM	1.01	0.88–1.16	0.89
25–49 KM	0.88	0.76–1.01	0.08
50–99 KM	0.77	0.65–0.91	0.002
≥100 KM	0.48	0.39–0.60	<0.001
Complete history/physical examination by a PCP			
No (referent)			
Yes	1.16	1.07–1.26	<0.001

likely to attend specialized clinics. Among the least marginalized survivors, 48 % attended at least one clinic compared with 36 % of the most marginalized group. In adjusted models, females were 20 % more likely than males to attend clinics, a phenomenon also observed in other health care contexts [29]. As anticipated, survivors diagnosed prior to the launch of the clinic in 1999 were 27 % less likely to attend a clinic than those diagnosed more recently. Although survivors can "self-refer" to these clinics, direct transition from a pediatric survivor clinic or active oncology clinic understandably leads to better attendance. Further, survivors who self-refer as adults usually do so when they have developed a symptomatic late effect or when functional limitations interfere with normal activity or employment. At this point, the opportunity for the clinic to provide health promotion or preemptive surveillance and intervention has been lost. Strategies to reach survivors who graduated from their pediatric center before the launch of the survivor clinics or who attended a survivor clinic but subsequently dropped out have not been implemented in Ontario. Such strategies might include public service announcements, particularly those that target subgroups at high risk for late effects (e.g., breast cancer after chest radiation; cardiomyopathy after anthracycline therapy). Another approach would be to contact survivors directly using current address information available in provincial databases. Although this may raise concerns about privacy, a recent initiative in Canadian province of British Columbia aims to contact approximately 3400 adult survivors who have been lost to follow-up.

Given the inconsistency with which survivors attend these clinics and the general discomfort among PCPs, empowerment of the survivor to seek appropriate care (regardless of care location or provider) is critical. However, survivor

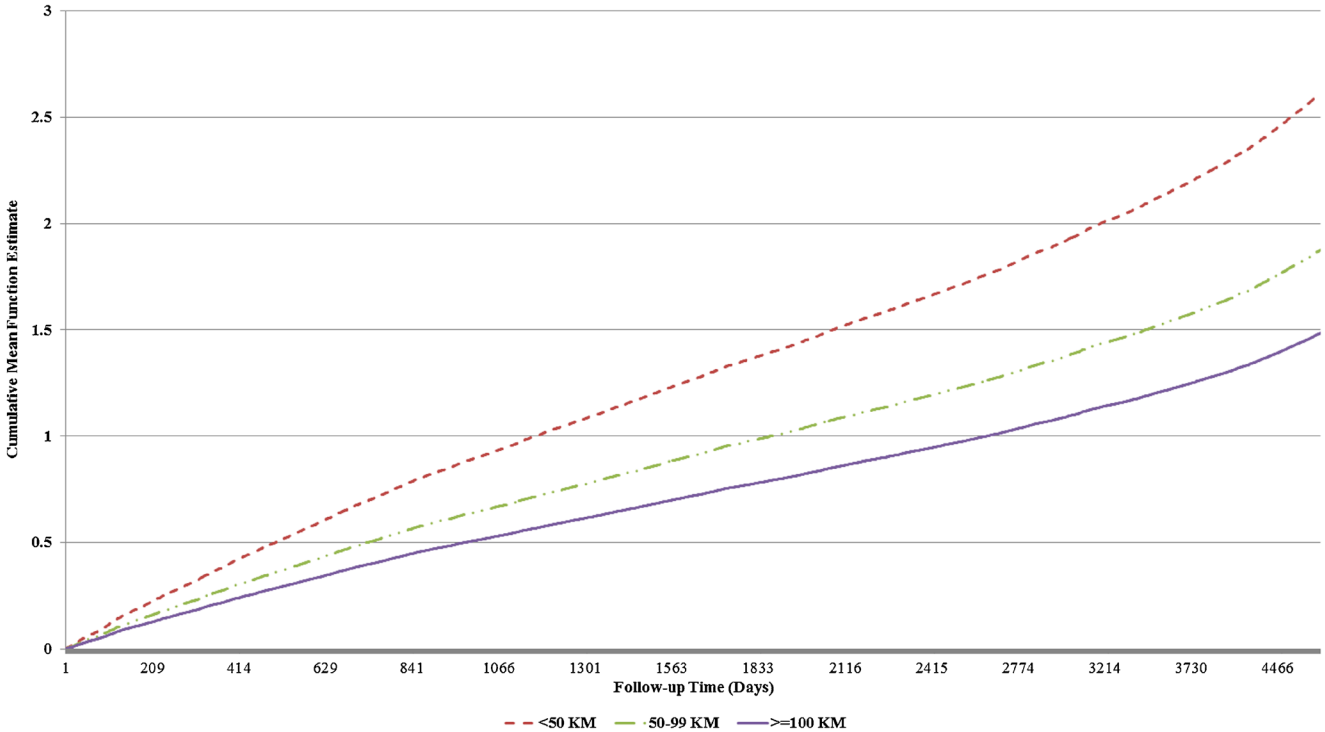

Fig. 2 Cumulative mean survivor clinic visits over time according to distance from nearest survivor clinic

knowledge about their diagnosis, treatment, and consequent risk for late effects is often inadequate [30, 31], creating a significant barrier to seeking appropriate care. Although provision of a treatment summary and survivor care plan that provides specific instructions regarding recommended long-term care and surveillance is now mandated by groups such as the American College of Surgeons Commission on Cancer [32], more focused interventions are necessary to ensure that survivors integrate these recommendations into their health-seeking behavior. For example, a randomized trial of the addition of a telephone encounter with an advanced practice nurse to a survivorship care plan demonstrated a twofold increase in compliance with cardiomyopathy screening in survivors treated with anthracycline chemotherapy [33]. Whether this improved compliance can be sustained over time and whether such interventions can be implemented in a cost-effective manner will require further research.

Several limitations must be considered when interpreting the results of this study. We did not assess survivor clinic attendance prior to age 18 years. However, attendance at specialized survivor clinics during childhood is almost 100 %, and consequently, we chose to focus on attendance as adults, when the risk of attrition rises. Since the outcomes were derived from administrative data and not direct patient assessment, we could not estimate the presence or absence of late effects that may drive care-seeking behavior. Knowledge of the types and intensity of cancer therapy allows for estimation of risk for late effects, but there is no way to know if survivors who visited the clinics had existing morbidities from their prior therapy. This study focuses on survivors in the most populous Canadian province where there is an existing network of survivor clinics and no cost for access, which may impact generalizability to other jurisdictions. However, the Ontario model serves as a paradigm for centering survivor care on a network of specialized clinics. Given that other countries have publicly funded health systems and many survivors in the USA have individual health insurance, the experience with attendance in the Ontario model should have direct relevance to survivor programs elsewhere. Finally, this study focuses on the patterns of attendance at specialized clinics and does not assess how clinic attendance impacts quality of care or long-term health outcomes. Ongoing research by our group is assessing the impact of clinic attendance on compliance with published surveillance guidelines and with the risk for requiring care in an emergency department.

In conclusion, a survivor care strategy that relies exclusively on attendance at a cancer center-based survivor clinic is unlikely to meet the needs of all adult survivors of childhood cancer. Models must be built that consider geographic distribution, the intensity of therapy and the risk for late effects, and access to PCPs willing to care for survivors. Developing tools and programs to facilitate the provision of care by PCPs and interventions to empower survivors to recognize their risks and to seek appropriate care will be critical for maximizing the long-term health outcomes of this growing and vulnerable population.

Compliance with ethical standards

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Conflict of interest Paul Nathan, Mohammad Agha, Jason Pole, Rinku Sutradhar, Astrid Guttmann, David Hodgson, and Mark Greenberg all declare that they have no conflict of interest

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Ethical approval was obtained from the Research Ethics Boards at all five pediatric cancer centers in the province of Ontario. Since the study involved database/registry research only and no patients were contacted as part of the study procedure, the ethics boards waived the need for informed consent.

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