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An investigation of survivorship clinic attendance among childhood cancer survivors living in a five-state rural region

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Abstract
Purpose Cancer survivorship clinics manage cancer-related health complications and are available primarily in urban areas. We examine how demographic, clinical, and geographic-based characteristics are associated with attendance at the only pediatric survivorship clinic in a largely rural, multistate region.
Methods One thousand eight hundred sixteen cancer survivors were diagnosed at age ≤ 25 from 1986 to 2005 while living in the region. Cox models incorporating death as a competing risk and generalized estimating equations calculated hazards ratios (HR) for characteristics measured at the clinic's opening. Subjects were followed from the clinic opening their first visit, death, emigration from the catchment area, or December 31, 2014.
Results Five percent of survivors visited the clinic. Attendance is positively associated with a leukemia or lymphoma diagnosis (HR = 3.32, 95% confidence interval [CI] = 1.72–6.78 vs CNS tumors), previous relapse (HR = 1.78, 95% CI = 1.00–3.19), and residing >100 mi from the clinic (HR = 2.05, 95% CI 1.03–4.10). Survivors aged ≥ 31 years at clinic opening (HR = 0.19, 95% CI = 0.07–0.54) are less likely to attend than younger survivors. Residence between 16 and 100 mi had an inverse association with attendance, although not significant.
Conclusion Survivorship clinics are not widely attended by survivors in this catchment region. Efforts should be made to recruit survivors aged ≥ 31 and diagnosed with CNS tumors. Distance has a complex association with attendance, which could be attributed to the limited availability of preventative services in regions > 100 mi from the clinic.
Implications for Cancer Survivors Survivors living in this catchment region may not be receiving care necessary to prevent severe late effects.

Keywords Survivorship · Pediatric oncology · Geography · Late effects

Introduction

Within 20 years of diagnosis, 60% of childhood cancer survivors report at least one serious condition that is related to their prior cancer or associated therapies [1]. Survivor-focused preventative care could reduce the severity of late health effects [2, 3], but survivorship care is commonly managed by primary care physicians or noncancer specialists [4–6]. In contrast, survivorship clinics are staffed by physicians and/or nurse specialists who diagnose and manage health conditions specific to childhood cancer survivors. Clinic attendance is linked to improvements in patient awareness of late effects, management of health conditions, improved mental health, and decreased emergency department visits [7–11].

The 60 survivorship clinics in the USA are largely located in urban areas [12]. The location of these clinics may pose a barrier to survivors living in large rural areas. In Ontario, Canada, 43% of childhood cancer survivors attend a survivorship clinic despite access to free clinic care, suggesting that indirect costs like

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time off work, travel time, or a lack of awareness of late effects may impact attendance [5]. In contrast, the survivorship clinic at the Yale HEROS Clinic in New Haven, Connecticut had a 37% attendance rate, with insurance being the primary predictor of attendance. Since both clinics are in urban areas, and the Yale cohort only included survivors living ≤ 100 mi of the clinic [13], more information about rural survivors in the USA is needed.

The largely rural Intermountain West is one of the largest regions in the continental USA, consisting of the states Utah, Idaho, Montana, Nevada, and Wyoming. Mining, fossil fuel extraction, agriculture, and tourism are major industries, with growing science and technology industries [14]. The region's population is growing faster than the national rate [15]. Intermountain states typically have a few dense metropolitan areas which are surrounded by rural and frontier areas with low population density [16]. In Utah, 75% of the population lives on 4.3% of its total landmass [17, 18]. The five-state region is served by one childhood oncology center located in Salt Lake City, Utah; nearly 25% of childhood cancer patients travel two or more hours for cancer treatment [19]. The oncology center is located within a larger university health sciences center. In 2010, a survivorship clinic on the same medical campus as the children's oncology center began treating adult survivors of childhood cancer and young adults who were diagnosed with common pediatric cancers. Since travel is a significant time investment, the burden of continued travel for posttreatment care may discourage rural populations from attending the survivorship clinic.

We characterize a retrospective cohort of childhood, adolescent, and young adult cancer survivors in the Intermountain West who were diagnosed and/or received at least part of their treatment at the region's only pediatric oncology center. We identify demographic, clinical, and geographic predictors of survivorship attendance among survivors who were eligible to attend the clinic and were living in the clinic's catchment area when it opened. Using a statewide population resource, the Utah Population Database [20], we created limited residential histories for survivors in the Intermountain West region to establish their residence at the time of the survivorship clinic opening.

Methods

Study population

Intermountain Health (IH) is a health system comprised of 185 clinics and 23 hospitals located throughout the Intermountain West. IH includes the only pediatric oncology center (Primary Children's Hospital (PCH)) in the region [21]. We obtained records for patients who were diagnosed with cancer at ≤ 25 years, or who were diagnosed with a pediatric cancer between 18 and 25 years at an IH facility, and were treated in an IH facility between January 1, 1986, and December 31, 2005. January 1, 1986 was the earliest date that cancer registry records were available for our study population. December 31, 2005 was the last date that a cancer patient could be diagnosed and be considered eligible for attendance. Patients who were diagnosed while living in Utah, Idaho, Montana, Nevada, or Wyoming were considered residents of our catchment region.

To qualify for the clinic, patients were at least 3 years off treatment and did not require surveillance imaging more than once a year. Patients could either be seen by the late effects medical director at PCH or at Huntsman Cancer Institute, which is located on the same medical campus at PCH. Treating oncologists were asked to refer patients to the clinic when they considered patients ready for survivorship care, which was typically 5 years postdiagnosis. Because of this continual following, we defined clinic eligibility in this study as 5 years postdiagnosis to ensure that patients were completely off therapy. When the clinic first opened, several methods were employed to advertise. The clinic was advertised on the Huntsman Cancer Institute's Wellness center website. Press releases were also distributed to local media resulting in newspaper articles advertising the clinic services. Finally, postcards were sent to all primary care providers (PCPs) in Utah and PCPs in the IH system located in surrounding states (Idaho, Montana, Nevada, and Wyoming).

Residential history

Residential addresses for participants were supplied by the Utah Population Database (UPDB), which links birth, death, driver licenses, voter registration, marriage, and divorce records to create a lifetime record of each Utah resident. UPDB is a dynamic database, with new records added continually. UPDB linked these records to IH cancer diagnosis, hospitalization, and surgical records that contain ZIP codes. These records provide UPDB with a first known date of residence in Utah, as well as a last known date of residence in the state. Individuals who completed any of the above records while in Utah were included in UPDB irrespective of state of birth or current state of residence [22], enabling UPDB to track individuals who do not live in Utah in a limited fashion. For non-Utahans, every cancer diagnosed at PCH, visit to an IH facility for cancer-related treatment or hospitalization, marriage, divorce, birth, or death that took place in Utah were recorded in UPDB. UPDB has an average of eight records per person in their database [23].

UPDB has constructed family trees and family residential histories using the aforementioned records. If a patient was aged less than 18 years at diagnosis, the mother's address information was recognized as the address of residence. Once a cancer case became 18 years or older, the patient's individual records were the source for residential addresses.

UPDB matched IH patients that we identified to residential records in their system. We received residential ZIP codes

affiliated with all UPDB records through December 31, 2015 for our cohort. Using these records, we identified patients' residence at diagnosis, at clinic opening, and during follow-up. We verified UPDB ZIP codes with IH records containing ZIP codes at diagnosis to ensure the records were in agreement.

Cohort description

Patients were included if they (1) were diagnosed between January 1, 1986, and December 31, 2005; (2) lived in Utah, Idaho, Montana, Nevada, or Wyoming at diagnosis; (3) lived in the catchment region 5 years postdiagnosis; and (4) resided in the catchment area as of July 31, 2010 according to their most recent UPDB record. We also limited inclusion to patients who had survived 5 years past their first diagnosis date to ensure their clinic eligibility.

Patients were followed from the clinic's opening in August 1, 2010 until their first clinic visit, death, emigration out of the catchment region as determined by UPDB records dated after August 1, 2010, or until the end of follow-up in December 31, 2014. Participants were censored if they did not visit the clinic and did not have a record in UPDB between August 1, 2010, and December 31, 2014.

Attendance was determined using the survivorship clinic patient registry, which contained 178 names and addresses of patients that attended the clinic through December 31, 2014. Attenders visited the clinic at least once. Patients were matched by first and last name to IH and UPDB records. We excluded 51 attenders that did not appear in IH or UPDB records; these patients visited the survivorship clinic but were diagnosed and treated out of the catchment area. We also excluded 42 attenders that were diagnosed from 2006 to 2009 because they did not meet the cohort inclusion criteria of being 5 years from diagnosis at the time of clinic opening.

Vital status for patients as of January 1, 2014 was determined in the following manner: (1) anyone that visited an IH clinic on or after January 1, 2014 was presumed to be alive as of January 1, 2014; and (2) vital status for patients without a clinic visit on or after January 1, 2014 was queried using the National Death Index (NDI) using first, middle, and last name, birthdate, sex, and birth state.

Demographic and clinic-related characteristics

UPDB supplied sex and birthdate. Ethnicity was ascertained through a two-step process: (1) ethnicity recorded by birth certificates, medical records, and driver licenses in UPDB records; and (2) a two-step surname-matching process [24]. Combining these methods has a specificity of 95% and sensitivity of 83% [25]. Cancer type was determined by aggregating the primary International Classification for Childhood Cancer codes provided by IH into three groups: (1) leukemia or lymphoma, (2) central nervous system tumors (CNS), and (3) solid tumors (soft tissue sarcomas, bone tumors, germ cell tumors, and retinoblastoma). Relapses that occurred before July 31, 2010 were recorded by IH and used as a dichotomous variable. Age at diagnosis was defined using IH cancer diagnosis records. Age at clinic opening was determined using UDPB birth records.

ZIP code characteristics

Distance to clinic

We obtained population-weighted centroids for every ZIP code tabulation area (ZCTA) in states in our catchment region [26]. Participants' most recent ZIP codes as of August 1, 2010 from UPDB records were linked to ZCTAs in ArcMap (match rate 99%). Unmatched ZIP codes ($n = 5$) were matched manually to the geographically closest ZCTA. We calculated the Euclidean distance between the population-weighted ZCTA centroid and the survivorship clinic in miles. Distance from the clinic was separated by geographic distances of ≤ 15, 16 to 50, 51 to 100, and > 100 mi.

Median household income

We downloaded ZCTA data from the 2010 US Census [27], including median annual household income for all ZCTAs [28]. These data were matched to participants by ZIP codes.

Statistical methods

We calculated chi-square tests and p values for differences in attendance between each characteristic. Individual and multivariable hazard ratios were calculated by Cox proportional hazards models that incorporated death as a competing risk and generalized estimating equations (GEE) to account for geographic clustering. Variables were included in the multivariable analysis if their individual analyses yielded nonnull hazards ratios. ZIP code distances to clinic and income were correlated so they were analyzed separately; the same approach was taken with age at clinic opening and age at diagnosis. We ran four models with different combinations of distance, income, and age variables. Model 1 includes age at diagnosis and distance from the clinic, model 2 includes age at diagnosis and median income, model 3 includes age at clinic opening and distance from the clinic, and model 4 includes clinic opening and median income. Longer distances were correlated with increasing rurality, so urban/rural area was not included in the model.

Prior to using the Cox models, we examined Kaplan-Meier and log-log curves and did not see departures from the proportional hazards assumption that concerned us, with the exception of year of diagnosis [29]. Because of this, we stratify our multivariable analyses by diagnosis year.

Results

A total of 1812 childhood, adolescent, and young adult cancer survivors met our eligibility criteria and resided in the five-state catchment area. Of the most recent records, 139 had non-Utah ZIP codes and 1677 had Utah ZIP codes. Eighty-eight percent of the most recent records in UPDB were dated between January 1, 2004, and July 31, 2010.

Most survivors were Non-Hispanic (90.1%) and diagnosed with leukemia and lymphoma (40.9%) (Table 1). Nearly a fourth of the cohort (17.0%) lived > 100 mi from the clinic when it opened. Over 65% lived in ZIP codes with median incomes of $50,000 to $90,000 when the clinic opened.

Table 1 Characteristics of childhood, adolescent, and young adult cancer survivors in a five-state rural region

		Overall (n = 1816)		Clinic nonattender (n = 1731)		Clinic attender (n = 85)		p value[a]
		N	%	N	Col %	N	Col %	
Demographic characteristics								
Sex	Female	806	44.4	763	44.1	43	50.6	0.24
	Male	1010	55.6	968	55.9	42	49.4	
Ethnicity	Hispanic	180	9.9	170	9.8	10	11.8	0.56
	Non-Hispanic	1636	90.1	1561	90.2	75	88.2	
Residential area	Rural	327	18.0	302	17.4	25	29.4	0.02
	Urban	1487	81.9	1427	82.4	60	70.6	
Clinic-related characteristics								
Type of cancer	Central nervous system tumors	471	25.9	460	26.6	11	12.9	< 0.01
	Leukemia and lymphoma	742	40.9	694	40.1	48	56.5	
	Solid tumors	603	33.2	577	33.3	26	30.6	
Previous relapse	No relapse	1655	91.1	1583	91.5	72	84.7	0.03
	Relapse	161	8.9	148	8.6	13	15.3	
Year of diagnosis	1986–1997	841	46.3	817	47.2	24	28.2	< 0.01
	1998–2005	975	53.7	914	52.8	61	71.8	
Time since diagnosis	5 years or less	136	7.5	130	7.5	6	7.1	0.01
	6 to 10 years	632	34.8	595	34.4	37	43.5	
	11 to 15 years	459	25.3	431	24.9	28	32.9	
	16 or more years	589	32.4	575	33.2	14	16.5	
Age at diagnosis	0 to 4 years	564	31.1	530	30.6	34	40.0	< 0.01
	5 to 10 years	360	19.8	337	19.5	23	27.1	
	11 to 17 years	463	25.5	439	25.4	24	28.2	
	18 to 25 years	429	23.6	425	24.6	4	4.7	
Age at clinic opening	10 years or less	168	9.3	160	9.2	8	9.4	< 0.01
	11 to 20 years	515	28.4	472	27.3	43	50.6	
	21 to 30 years	662	36.5	633	36.6	29	34.1	
	31 years or older	471	25.9	466	26.9	5	5.9	
Area-level characteristics								
Residential distance from the clinic[a]	≤ 15 mi	565	31.2	540	31.3	25	29.8	0.04
	15 to 50 mi	728	40.2	697	40.4	31	36.9	
	51 to 100 mi	211	11.7	206	11.9	5	6.0	
	> 100 mi	307	17.0	284	16.4	23	27.4	
Area-level median income[b]	≤ $30 to $50,000	562	31.0	534	30.9	28	33.3	0.5
	$51K to $70K	847	46.8	812	47.0	35	41.7	
	$71 to $90K	351	19.4	331	19.2	20	23.8	
	> $91,000	50	2.8	49	2.8	1	1.2	

[a] p values compare nonattenders to attenders

[b] Determined by ZIP code tabulation area

Survivors were, on average, age 21 years at cohort entry, diagnosed at age 11 years, and followed for 4 years.

Of the 1812 survivors, only 5% attended the clinic. Compared to nonattenders, attenders were diagnosed with cancer at a younger age (10.8 vs 8.5 years, $p < 0.01$). Leukemia and lymphoma survivors were more likely to attend the clinic (6.5 vs 2.3% of CNS and 4.3% of solid tumors, $p < 0.01$). Attendance for survivors diagnosed between 1986 and 1997 differed from those diagnosed from 1998 to 2005 (2.9 vs 6.3%, $p < 0.01$).

Table 2 shows similar effect estimates and statistical significance for sex, ethnicity, and cancer type for models considering age at diagnosis and age at clinic opening entry separately. All estimates for attendance for diagnosis of leukemia and lymphoma and solid tumors were significantly higher compared to CNS tumors. Relapses that occurred before the clinic opening had a positive, significant association with clinic attendance in both models. Males were less likely to attend, but the estimate is not significant.

Models 1 and 2 and models 2 and 3 yield similar results for age at diagnosis and clinic opening entry, respectively. Effect estimates for distance and income for models 1 and 3 and models 2 and 4 also yield similar results. As such, we report results from models 1 and 3. In models 1 and 2, diagnosis at age 18 to 25 years was significantly lower than diagnosis at age ≤4 years (model 1: HR = 0.15, 95%

Table 2 Combined hazards ratios for late effects clinic attendance among childhood, adolescent, and young adult cancer survivors in a five-state rural region

		Age at diagnosis				Age at clinic opening			
		Model 1: distance		Model 2: income		Model 3: distance		Model 4: income	
		N = 1811		*N* = 1810		*N* = 1811		*N* = 1810	
		HR	95% CI	HR	95% CI	HR	95% CI	HR	95% CI
Demographic characteristics									
Sex	Female	Ref		Ref		Ref			
	Male	0.78	0.53–1.15	0.74	0.50–1.10	0.78	0.53–1.16	0.75	0.51–1.11
Ethnicity	Hispanic	Ref		Ref		Ref			
	Non-Hispanic	0.82	0.41–1.66	0.86	0.43–1.69	0.87	0.43–1.75	0.94	0.48–1.85
Clinic-related characteristics									
Type of cancer	Central nervous system tumors	Ref		Ref		Ref			
	Leukemia and lymphoma	3.32*	1.72–6.78	3.39*	1.68–6.82	3.30*	1.65–6.59	3.29*	1.62–6.69
	Solid tumors	2.55*	1.19–5.47	2.50*	1.15–5.43	2.31*	1.10–4.85	2.30*	1.08–4.90
Age at diagnosis	0 to 4 years	Ref		Ref					
	5 to 10 years	1.12	0.59–2.13	1.15	0.60–2.21				
	11 to 17 years	0.87	0.50–1.52	0.92	0.54–1.58				
	18 to 25 years	0.15*	0.05–0.42	0.16*	0.06–0.44				
Age at clinic opening	10 years or less					Ref			
	11 to 20 years					1.52	0.71–3.28	1.50	0.69–3.25
	21 to 30 years					0.82	0.37–1.81	0.82	0.37–1.83
	31 years or older					0.19*	0.07–0.54	0.19*	0.07–0.53
Previous relapse	No relapse	Ref		Ref		Ref			
	Relapse	1.78*	1.00–3.19	1.82*	1.01–3.29	1.96*	1.11–3.45	1.94*	1.09–3.47
ZIP code characteristics									
Distance from the clinic	≤ 15 mi	Ref				Ref			
	15 to 50 mi	0.95	0.53–1.70			0.93	0.52–1.66		
	51 to 100 mi	0.59	0.24–1.50			0.59	0.24–1.46		
	> 100 mi	2.05*	1.03–4.10			2.05*	1.02–4.09		
Median income, by ZCTA	> $91,000			Ref				Ref	
	$71 to $90K			2.50	0.29–21.7			2.56	0.31–21.16
	$51K to $70K			1.79	0.21–15.5			1.90	0.23–15.66
	≤ $30 to $50,000			2.17	0.25–19.0			2.34	0.28–19.32

* Significant at $p \leq 0.05$

CI = 0.05–0.42). In models 3 and 4, survivors aged ≥ 31 years at clinic opening were significantly less likely to attend the clinic than survivors aged ≤ 10 years at clinic opening (model 3: HR = 0.19, 95% CI = 0.07–0.54). Living > 100 mi from the clinic was significantly associated with attendance compared to living ≤ 15 mi from the clinic (model 1: HR = 2.05, 95% CI = 1.03–4.10). Survivors living 15 to 50 mi (model 1: HR = 0.95, 95% CI = 0.53–1.70) and 51 to 100 mi (model 1: HR = 0.59, 95% CI = 0.24–1.50) are less likely to attend than those living at closer distances, but not significant. Survivors living in ZIP codes with median incomes of $30,000 to $50,000 were more likely to attend the clinic than survivors living in ZIP codes of $91,000 or more, although the estimate is not significant (model 3: HR = 2.17, 95% CI = 0.25–19.0).

Analyses stratified by diagnosis year (Tables 3 and 4) show different trends. In the analyses with age at diagnosis (Table 3), leukemia and lymphoma (HR = 4.64, 95% CI = 1.84–11.74) and solid tumors (HR = 3.97, 95% CI = 1.47–10.68) are not significant for survivors diagnosed between 1986 and 1997 but are significant for survivors diagnosed between 1998 and 2005. Lower odds of attendance for survivors aged 18 and 25 years at diagnosis are significant for survivors diagnosed between 1986 and 1997 (HR = 0.11, 95% CI = 0.01–0.82) and between 1998 and 2005 (HR = 0.18, 95% CI = 0.05–0.61). For survivors diagnosed between 1986 and 1997, relapses (HR = 3.14, 95% CI = 1.26–7.86) are a significant predictor of attendance, but not for survivors diagnosed earlier.

For analyses with age at clinic opening (Table 4), survivors who were diagnosed between 1986 and 1997 and age ≥ 31 years at clinic opening are significantly less likely to visit the clinic than survivors diagnosed in the same time period and who were 11 to 20 years at clinic opening (model 3: HR = 0.19, 95% CI = 0.07–0.54). Leukemia and lymphoma and solid tumors are significant predictors of attendance for survivors diagnosed between 1998 and 2005, but not earlier

Table 3 Combined hazards ratios by year of diagnosis for late effects clinic attendance among childhood, adolescent, and young adult cancer survivors in a five-state rural region

	1986–1997 $n = 840$		1998–2005 $n = 971$	
	Hazard ratio	95% CI	Hazard ratio	95% CI
Demographic characteristics				
Sex				
Female	Ref		Ref	
Male	0.53	0.20–1.42	0.90	0.57–1.43
Ethnicity				
Hispanic	Ref		Ref	
Non-Hispanic	1.24	0.34–4.47	0.81	0.35–1.86
Clinical characteristics				
Type of cancer				
Central nervous system tumors	Ref		Ref	
Leukemia and lymphoma	1.94	0.66–5.72	4.64*	1.84–11.74
Solid tumors	0.88	0.26–2.97	3.97*	1.47–10.68
Age at diagnosis				
0 to 4 years	Ref		Ref	
5 to 10 years	0.40	0.11–1.40	1.71	0.80–3.68
11 to 17 years	0.38	0.12–1.17	1.30	0.67–2.53
18 to 25 years	0.11*	0.01–0.82	0.18*	0.05–0.61
Previous relapse				
No relapse	Ref		Ref	
Relapse	3.14*	1.26–7.86	1.17	0.45–3.05
ZIP code-level characteristics				
Distance from the clinic				
≤ 15 mi	Ref		Ref	
16 to 50 mi	1.15	0.50–2.65	0.90	0.43–1.85
51 to 100 mi	0.45	0.09–2.32	0.63	0.20–1.94
> 100 mi	2.05	0.73–5.76	1.70	0.75–3.86

* Significant at $p \leq 0.05$

Table 4 Combined hazards ratios by year of diagnosis for late effects clinic attendance among childhood, adolescent, and young adult cancer survivors in a five-state rural region, including current age

	1986–1997 $n = 840$		1998–2005 $n = 971$	
	Hazard ratio	95% CI	Hazard ratio	95% CI
Demographic characteristics				
Sex				
Female	Ref		Ref	
Male	0.53	0.21–1.34	0.92	0.59–1.46
Ethnicity				
Hispanic	Ref		Ref	
Non-Hispanic	1.23	0.30–5.11	0.82	0.37–1.80
Clinical characteristics				
Type of cancer				
Central nervous system tumors	Ref		Ref	
Leukemia and lymphoma	1.99	0.69–5.75	4.36*	1.75–10.88
Solid tumors	0.86	0.25–2.98	3.63*	1.36–9.71
Age at clinic opening[a]				
10 years or less			Ref	
11 to 20 years	Ref		1.61	0.73–3.59
21 to 30 years	0.45	0.16–1.30	0.96	0.41–2.27
31 years or older	0.22*	0.07–0.67		
Previous relapse				
No relapse	Ref		Ref	
Relapse	3.72*	1.67–8.28	1.19	0.48–2.94
ZIP code-level characteristics				
Distance from the clinic				
≤ 15 mi	Ref		Ref	
16 to 50 mi	1.11	0.47–2.62	0.87	0.42–1.80
51 to 100 mi	0.47	0.10–2.23	0.65	0.22–1.92
> 100 mi	2.14	0.74–6.15	1.86	0.83–4.16

* Significant at $p \leq 0.05$

[a] No participants in 1986–1997 are aged 10 years or younger, and no participants in 1998–2005 are aged 31 years or younger

years. Relapses are also significant in survivors diagnosed in the earlier time period.

Discussion

In our assessment of pediatric cancer late effects clinic attendance in the Intermountain West region, we found that 5% of survivors in the IH system and living the five-state catchment region visited the survivorship clinic. Our attendance rate is substantially lower than those of other studies, which ranged from 27.8% attendance at Yale University in New Haven [13] to 85% at St. Jude Children's Research Hospital [30]. These cohorts and our cohort differ significantly in terms of insurance accessibility, geographic scope, and protocol involved with clinic scheduling and follow-up. However, we found similar patterns in attendance by age at clinic opening entry as previous studies, with older survivors much less likely to attend the clinic than younger survivors [5, 13, 30].

Previous studies of childhood cancer survivors aged 30 years or older found that they are less likely to seek preventative care than younger survivors [1, 31]. Since the risk and severity of late effects worsen with age, ensuring that older survivors have access to preventative services is critical maintaining their health. Future research should investigate the role of time constraints related to family or career responsibilities, awareness of late effects [7, 32], health insurance, or cost of care on the reduced utilization of survivorship care by age [33]. Innovative strategies to reach older survivors in the Intermountain West through advertising or primary care providers may be needed to ensure they are aware of survivorship services.

Insurance coverage is a possible explanation for the differences in attendance between our study and earlier reports. In Ontario, 43% of the patients had attended their clinic that also

serves a large geographic region; however, all Ontario residents had access to health insurance that provided free care at the survivorship clinic [5]. This is much higher than the 5% attendance rate in our study. In the USA, the prevalence of insurance coverage among adults aged 18 and older increases with income [34]. Among cancer survivors, survivors aged 25 to 39 years are significantly more likely to experience a period where they do not have insurance coverage after diagnosis [35]. While we did not have information to examine individual health insurance coverage or income, our study did overlap with the rollout of the Patient Protection and Affordable Care Act, which expanded dependent coverage and had potential to improve insurance availability for survivors [36]. With potential upcoming changes in insurance availability with the new administration, longitudinal studies are needed to evaluate how changes in insurance and income affect late effects clinic attendance.

Our cohort and the St. Jude's cohort report that survivors who travel the longest distances are more likely to attend than those living a closer distance. Patients in the St. Jude's cohort lived a median of 194 mi from the clinic; attenders lived a median of 183 mi from St. Jude's, while nonattenders lived a median of 23.5 mi [30]. Similarly, survivors in our cohort who lived the farthest distance (> 100 mi) were significantly more likely to attend than those living ≤ 15 mi of the clinic. In contrast, the Yale cohort only included survivors living ≤ 100 mi of the clinic at 5 years postdiagnosis and found that travel time was not a significant predictor of clinic attendance [13]. While we did not examine travel patterns for patients in our cohort during their cancer treatment [19], many childhood cancer patients travel a substantial distance for oncology care in our catchment area, suggesting that certain patients may be used to traveling far distances and comfortable traveling for late effects care.

In addition, the relationship between clinic attendance and travel distance may be affected by the availability of primary care providers and physician referrals in certain geographic areas. The largest urban centers in the Intermountain West are located within 100 mi of the survivorship clinic, with the greatest density of primary care providers located in the urban areas surrounding Salt Lake County in Utah [37]. Outside of these urban areas are more sparsely populated frontier and rural areas with fewer healthcare providers. As such, providers living greater distances from the survivorship clinic may be more likely to refer patients to the clinic than those living closer to large urban centers, and patients may be more willing to travel to receive the care they prefer. Also, local patients may perceive that attending a specialized survivorship clinic is less convenient or not necessary because of the availability of local providers. Further research is needed to better understand travel patterns and geographic barriers to survivorship care.

Despite evaluating patients up to age 25 treated in the IH system, which includes the only children's hospital in our region, we found that 51 clinic attenders were not previous IH patients. When we include the 42 additional patients who did not meet eligibility criteria, our overall attendance rate increases to 9.8% but is still much lower than previous studies. Also, we found that attendance among survivors diagnosed from 1986 to 1997 was lower than survivors diagnosed from 1998 to 2005. It is likely that longer term survivors are less aware of the clinic services. Still, several patients diagnosed in these earlier years did attend the clinic, suggesting that outreach efforts (e.g., postcards to primary care providers) may have raised awareness for some patients. However, in general, the low attendance suggests greater efforts need to be made to expand referrals for longer term patients.

Our findings demonstrate the need to systematically refer patients to survivorship care in both urban and rural healthcare systems. When the survivorship clinic first opened, primary oncologists provided posttherapy care and were in charge of referring patients to the clinic when the provider decided the patient was ready. During this time, follow-up from the primary oncologist could last far longer than the usual 5 years posttherapy, which would replace survivorship clinic care. In addition, similar to previous studies, we report lower attendance among CNS tumor survivors. Because they are complex patients, CNS tumor survivors may be referred to other subspecialists or clinics rather than a survivorship clinic [38]. At PCH, a multidisciplinary clinic for CNS survivors was in operation when the survivorship clinic opened, but closed during our study time period. As the CNS survivors are not systematically being seen at the survivorship clinic, follow-up of these CNS survivors is needed to ensure they are receiving the necessary follow-up care.

Limitations include the lack of information on insurance and individual income which should be investigated in future studies [13]. In addition, follow-up for IH patients living outside of Utah is limited and so misclassification of residential address may have occurred. However, the addresses for non-Utah subjects were fairly consistent before and after the clinic opening, so we do not anticipate a large change in the addresses of IH patients living in our catchment region although emigration could still be a limitation. Since treatment data for survivors diagnosed from 1986 to 1997 was not readily available, future studies should examine attendance by the type, intensity, and duration of treatment. Because of the low attendance, this study may be underpowered to identify significant associations between individual- and area-level characteristics. Future studies should provide clarity regarding the role of insurance and type of treatment on survivorship clinic attendance. Additional work on clarifying the role of survivorship clinics in a complex healthcare system with multiple preventative care providers is necessary.

This is the first report on survivorship clinic attendance in a large, majority rural region and explored the role of distance on clinic attendance for survivors living in geographically

remote areas. The vast majority of childhood cancer survivors in the Intermountain West do not attend the survivorship clinic, and other rural areas may be similar. Our results indicate that additional outreach may be needed for survivors who are male, aged 31 or older, and who were diagnosed with solid tumors or CNS and other neurologic tumors. Efforts to contact survivors living within 100 mi of the clinic need to be made. Clarifying the role of survivorship clinics is needed, given the complexity of the healthcare system and overlapping physician roles in survivorship care. Survivorship clinics have great potential to reduce the severity of late effects among cancer survivors and prevent serious conditions. Ensuring proper outreach, patient access, and clarification of the role of these clinics in the healthcare system should be of continued interest.

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Compliance with ethical standards

Conflict of interest The authors 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.

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