

## Geography and the Burden of Care in Pediatric Cancers

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**Background.** Childhood cancers typically require rigorous treatment at specialized centers in urban areas, which can create substantial challenges for families residing in remote communities. We evaluated the impact of residence and travel time on the burden of care for families of childhood cancer patients. **Procedure.** We conducted a cross-sectional, self-administered survey of 354 caregivers of pediatric cancer patients at a children's hospital serving a seven state area. Measures included the impact of cancer treatment on relocation, employment, schooling, and finances. We evaluated these domains by rural/urban residence and travel time (>1 hour and >2 hours) to the hospital in multivariable regression models. **Results.** Of the 29% of caregivers who reported moving residences as their child was diagnosed, 33% reported that the move was due to their

child's cancer. Rural and remote (e.g., >1 hour travel time) caregivers missed more days of work during the first month after diagnosis than did urban and local caregivers, however, these differences did not persist over the first 6 months of therapy. One-third of caregivers reported quitting or changing jobs as a direct result of their child being diagnosed with cancer. Rural respondents had greater out-of-pocket travel expenses and reported a significantly greater perceived financial burden. Rural patients missed more school days and were at an increased risk of having to repeat a grade. **Conclusions.** Childhood cancer has an appreciable impact on the lives of patients and caregivers. The burden is greater for those living far from a treatment center. *Pediatr Blood Cancer* 2014;61:1918–1924.

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### INTRODUCTION

Childhood cancers typically require treatment at specialized tertiary care centers that offer dedicated pediatric services and a multidisciplinary approach to care. Such specialized centers tend to be located in large urban areas and treat patients that come from diverse geographic areas and, often, great distances. Approximately, one-fifth of Americans live in rural areas [1] and many have travel times of greater than 1 hour to a designated National Cancer Institute Designated Cancer Center and other specialized treatment facilities [2]. Studies have shown that balancing a rigorous treatment schedule with school, work, and family obligations can be financially taxing for families affected by pediatric cancer [3–9].

The burden associated with childhood cancer care may be greater for families from rural communities and for those traveling great distances to primary cancer centers for therapy [10]. While there are several descriptive studies that assess the burden experienced by families during childhood cancer treatment [6,11–15], little is known about the effects of place of residence or travel time to therapy on that burden in the United States. Further, a more comprehensive understanding of burdens beyond the direct economic effects from therapy, such as the impact on parental employment and patient education, or from treatment-related factors such as emergency medical transport, is needed to better meet the supportive care needs for families. In the current study, we surveyed caretakers of pediatric cancer patients treated at a tertiary care center about the impact of cancer and its treatment on relocation, employment, schooling, and finances. Our analyses focused on determining whether residence (rural vs. urban) and travel time to the treatment center (>1 hour and >2 hours) imparts a greater care burden for these families.

### METHODS

#### Study Center

The study setting was the oncology clinic at Primary Children's hospital (PCH) in Salt Lake City. PCH serves as the

primary pediatric tertiary care center for one of the largest areas in the continental United States served by a single children's hospital.

#### Participant Sample

**Primary caretaker survey sample.** All primary caretakers of patients, diagnosed with a cancer between 0 and 18 years of age, who were at least 3 months from diagnosis, who were less than 60 months from the end of therapy, and who received care in the oncology clinic from January 1, 2010 to December 31, 2012 were eligible for participation. Of 449 caregivers who approached to participate during this timeframe, 356 completed the surveys. Two surveys were excluded due to a lack of travel time and rurality data, leaving 354 evaluable surveys (308 English and 46 Spanish language) for a response rate of 79%. To assure sample representativeness, we compared participant demographics to

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those for all outpatient visits using medical records for the same 2-year period.

## Survey Methods

Institutional Review Board approval was obtained. The burden of care, from the perspective of the patients' primary caregiver, was evaluated using a 48-item self-administered survey. Outcomes of interest included financial burden (travel costs, relocations, missed work, missed school, and perception of financial burden) and treatment-related factors, including utilization of medical transport. The survey was available in English and Spanish.

Both rurality and travel time were used as independent variables to allow for differential assessment of the impact of having easier access to the primary treatment center as well as having access to other resources more commonly found in urban areas. Rural versus urban residence at diagnosis was classified by Rural-Urban Commuting Area (RUCA) Codes [16,17], a ZIP code-based geographic taxonomy utilizing the Bureau of Census urban area and urban cluster definitions and work commuting flows to characterize areas regarding their rural and urban status.

Travel times were calculated using MapQuest. Travel time of greater/equal to or less than 1 hour from respondents' residence to PCH was categorized because this time cut off is used to direct patients with neutropenic fever to local hospitals, rather than PCH, for urgent care. Travel time of greater/equal to or less than 2 hours to PCH was categorized because this travel time encompasses the major metropolitan areas in the immediate region of the primary treatment center.

Cancer diagnoses were grouped into acute lymphoblastic leukemia (ALL), acute myeloid leukemia (AML), brain tumors, and solid tumors, creating groups with similar treatment schedules.

## Statistical Analysis

For comparison of the geographic groups on patient demographics, two-sample *t*-tests for continuous variables, Wilcoxon–Mann–Whitney tests for ordered categorical variables, and chi-square or Fisher's exact test, as appropriate, for unordered categorical variables were used. For dichotomous study outcomes, a multivariable Poisson regression was used, using time since diagnosis as the time variable in rate estimates. For continuous study outcomes, a multivariable linear regression was used adjusting for time since diagnosis. To compare groups for the ordered categorical variable "number of work days missed for each clinic visit," a multivariable logistic regression model was fitted using the rural/urban and travel time groups as the outcome variables and "days missed" categories as a continuous predictor variable, thus utilizing a linear trend test, as advocated by Woodward [18].

Multivariable models included all variables with *P*-value < 0.20 for association with urban/rural residence or travel time from univariate analysis (Table I). For all multivariable models, there were sufficient sample sizes and sufficient number of events to avoid over-fitting. The final models were adjusted for age at diagnosis, language, race, ethnicity, education, religion, and incorporated time since diagnosis. Missing data was not imputed. Rather, list-wise deletion was utilized (subjects with missing data were excluded from analyses for which the missing data were part of the model). All reported *P*-values are two sided. Statistical analyses were performed using Stata (College Station, TX).

## RESULTS

### Caregiver/Patient Characteristics

The mean age of patients were 6.7 years (SD 5.4, median 5, IQR 2,11) at cancer diagnosis and 8.8 years (SD 5.6, median 7.5, IQR 4,13) at the time of the survey, with a mean time from diagnosis to survey of 2.2 years (SD 2.2, median 1.5, IQR 0.4,3.1). The most common cancer diagnosis was ALL (44%), followed by solid tumors (37%), brain tumors (13%), and AML (5%) (Table I). Respondents did not differ significantly from those who refused participation in terms of age, gender, ethnicity, cancer, or time from diagnosis. A review of all outpatient medical records over the same 2-year period found that the surveyed cohort is generally representative of our patient population (36% with ALL diagnosis, 54% male, 13.1% Hispanic, a mean age of 9.7 years, and 27% of patients traveling >2 hours to the primary treatment center).

### Travel and Transportation

Rural dwellers accounted for 18.6% of the respondents (Table I). Mean travel time from patients' homes to the treatment center was 110 (SD 167, median 52, IQR 29,131) minutes with 39% and 26% reporting >1 and >2 hours travel times, respectively. Over 98% of caregivers utilized a car as transportation for routine clinical visits and hospitalizations, while a small fraction used a taxi (0.8%), train (3.3%), bus (0.5%), or airplane (1.7%).

There was no difference in the proportion of rural versus urban respondents whose child required a transfer by ambulance at any time since their cancer diagnosis. However, travel of >1 hour was found to increase the risk of requiring an ambulance (unadjusted percents 30% for >1 hour vs. 20% for <1 hour, aRR 1.6, 95% CI 1.1 to 2.6, *P* = 0.03). Likewise, a travel time of >2 hours increased the risk of ambulance utilization (unadjusted percents, 28% vs. 23%, respectively), although this was marginally significant in adjusted models (aRR 1.6, 95% CI 1.0 to 2.6, *P* = 0.06). Rurality was strongly associated with increased utilization of emergency air (airplane/helicopter) transport with 23% of rural respondents reported utilizing air transport in contrast to 5% of urban respondents (aRR 5.9, 95% CI 2.7 to 13.1, *P* < 0.001). This difference was partially explained by the effect of increasing travel time as a travel time of >1 hour was associated with an increased risk of emergency air transport (unadjusted percents: 15% for >1 hour vs. 4% for <1 hour, aRR 3.8, 95% CI 1.7 to 8.5, *P* = 0.001). Likewise, 21% of those traveling >2 hours versus 4% for <2 hours, utilized emergency air transport (aRR 7.2, 95% CI 3.5 to 14.5, *P* < 0.001) (Table II).

More rural patients (66%) received some type of healthcare outside of PCH at least one time after their diagnosis compared to urban patients (46%). Likewise, 70% of patients traveling >2 hours, compared to 43% for <2 hours, received care at another facility.

### Relocation of Residence

Overall, 29% of caregivers reported moving residences as their child was diagnosed with cancer. Of these, 33% reported that the move was directly due to their child having cancer, citing "being closer to the treatment center" and "being closer to family support" as the most common reasons. Greater travel time increased the likelihood of the move being "directly due to cancer" (aRR1.7, *P* = 0.2 for travel time >1 hour, and RR 2.1, *P* = 0.05 for travel time >2 hours) (Table II).

**TABLE I. Characteristics of Childhood Cancer Patients and Caregivers by Urban/Rural Residence and by Travel Time to Treatment Center**

	Residence <sup>a</sup>		Travel time to treatment center		Travel time to treatment center		P-value <sup>b</sup>	
	Overall [n = 354]	Rural (18.6%) [n = 66]	Urban (81.4%) [n = 288]	≥1 hour (39.3%) [n = 139]	<1 hour (60.7%) [n = 215]	≥2 hours (25.7%) [n = 91]		<2 hours (74.3%) [n = 263]
Age of cancer patient, yrs, mean ± SD	8.8 ± 5.6	9.6 ± 6.2	8.7 ± 5.5	9.2 ± 6.0	8.6 ± 5.4	9.4 ± 6.1	8.6 ± 5.5	0.27
Time from dx to survey-yrs, mean ± SD	2.2 ± 2.2	2.0 ± 1.8	2.2 ± 2.3	2.2 ± 2.3	2.1 ± 2.2	2.0 ± 1.9	2.2 ± 2.3	0.49
Male (patient), n(%)	195 (56)	36 (55)	159 (57)	79 (57)	116 (56)	50 (56)	145 (57)	0.86
Travel time, min, mean ± SD	110 ± 167	212 ± 104	87 ± 169	225 ± 220	35.8 ± 13.9	300 ± 241	44.5 ± 23.7	<0.01
Hispanic ethnicity, n (%)	71 (20)	11 (17)	60 (21)	22 (16)	49 (23)	52 (20)	19 (21)	0.86
White race, n (%)	292 (82)	59 (89)	233 (81)	121 (87)	171 (80)	76 (84)	216 (82)	0.76
Cancer type, (n%)								0.002 <sup>b</sup>
Acute lymphoblastic leukemia (ALL)	157 (44)	30 (45)	127 (44)	58 (42)	99 (46)	35 (38)	122 (46)	
Acute myeloid leukemia (AML)	18 (5)	3 (5)	15 (5)	6 (4)	12 (6)	3 (3)	15 (6)	
Brain tumor	45 (13)	2 (3)	43 (15)	12 (9)	33 (16)	5 (5)	40 (15)	
Solid tumor	132 (37)	31 (47)	101 (35)	63 (45)	69 (32)	48 (52)	84 (32)	
Primary language (caregiver)								
English, n (%)	308 (87)	62 (94)	246 (85)	128 (92)	180 (84)	81 (89)	227 (86)	0.51
Spanish, n (%)	46 (13)	4 (6)	42 (15)	11 (8)	35 (16)	10 (11)	36 (13)	0.82
Household income								
Category, n (%)								
<\$19,999	44 (13)	5 (8)	39 (15)	18 (14)	26 (13)	12 (14)	32 (13)	
\$20,000–\$39,000	82 (25)	21 (34)	61 (23)	30 (23)	52 (27)	22 (26)	60 (25)	
\$40,000–\$59,000	71 (22)	12 (20)	59 (22)	33 (26)	38 (19)	18 (21)	53 (22)	
\$60,000–\$79,000	52 (16)	11 (18)	41 (16)	19 (15)	33 (17)	13 (15)	39 (16)	
\$80,000–\$99,000	41 (13)	9 (15)	32 (12)	17 (13)	24 (12)	13 (15)	28 (12)	
Over \$100,000	35 (11)	3 (5)	32 (12)	12 (9)	23 (12)	6 (7)	29 (12)	0.85 <sup>c</sup>
Preferred not to answer <sup>c</sup> , n	29	5	24	10	19	7	22	0.59
Highest level of education of caregiver (%)								
Grades 1 through 8	23 (7)	2 (3)	21 (7)	4 (3)	19 (9)	4 (5)	19 (7)	
Grades 9 through 11 (some high school)	16 (5)	3 (5)	13 (5)	5 (4)	11 (5)	5 (6)	11 (4)	
High school or GED	43 (12)	10 (16)	33 (12)	23 (17)	20 (10)	13 (15)	30 (12)	
Some college, associate degree or vocational tech school	116 (34)	26 (42)	90 (32)	45 (34)	71 (34)	32 (37)	84 (33)	
College graduate or more	146 (42)	21 (34)	125 (44)	57 (43)	89 (42)	32 (37)	114 (44)	0.09 <sup>c</sup>
Preferred not to answer <sup>c</sup> , n	10	4	6	5	5	5	5	0.71
Insurance status (patient), n (%) <sup>d</sup>								
No insurance	6 (2)	1 (2)	5 (2)	2 (1)	4 (2)	2 (2)	4 (2)	0.67
Medicaid insured	128 (36)	25 (38)	103 (36)	52 (37)	76 (35)	33 (36)	95 (36)	0.98

TABLE I. (Continued)

	Residence <sup>a</sup>		Travel time to treatment center		Travel time to treatment center		P-value <sup>b</sup>
	Rural (18.6%) [n = 66]	Urban (81.4%) [n = 288]	≥1 hour (39.3%) [n = 139]	<1 hour (60.7%) [n = 215]	≥2 hours (25.7%) [n = 91]	<2 hours (74.3%) [n = 263]	
Religion of caregiver (%)							
Religion LDS	218 (63)	185 (66)	83 (61)	135 (64)	50 (56)	168 (65)	0.21
Religion catholic	53 (15)	40 (14)	19 (14)	34 (16)	15 (17)	38 (15)	0.13 <sup>b</sup>
Religion other	41 (12)	30 (11)	22 (16)	19 (9)	16 (18)	25 (10)	
Not religious, n (%)	35 (10)	27 (10)	13 (9)	22 (10)	8 (9)	27 (11)	
Religious service attendance frequency (caregiver)							0.62
							0.03 <sup>b</sup>

<sup>a</sup>Rural/urban residence was based on Rural-Urban Computing Area (RUCA) codes at the zip code level. RUCA codes were developed by the University of Washington Rural Health Research Center and the USDA Economic Research Service (ERS), with the support of the federal Health Resource and Service Administration's Office of Rural Health Policy and the ERS using standard Census Bureau urbanized area and urban cluster definitions in combination with work commuting data to characterize census tracts and later zip codes. The 10 RUCA categories were aggregated into urban (1-3) and rural (4-10) as recommended by the WWAMI (Washington, Wyoming, Alaska, Montana, and Idaho) Rural Health Research Center. <sup>b</sup>P-values reflect the significance of the differences between comparison groups with respect to each variable. P-Values for variables with >2 options (i.e., cancer type, income, education, religion) reflect the differences in distribution among the comparison groups of interest. Variables found to differ among comparison groups with a P-value of <0.2 were included in multivariable models. <sup>c</sup>P-values reflect the significance of the differences between comparison groups with respect to number of subjects with missing data. Missing data includes the following: Age (n = 6), Gender (n = 8), Ethnicity (n = 2), Income (n = 29), Education (n = 10), Religion (n = 7), Health insurance (1). <sup>d</sup>All patients not listed as Medicaid insured or uninsured had private or military insurance.

### Employment/Financial Burden

Caregivers missed a mean of 17.3 days (SD 32, median 12, IQR 7,20) of work during the first month after diagnosis, with rural families missing significantly more days than urban respondents (24 vs. 16 days, respectively, adjusted mean difference 10.1, 95% CI 0.66 to 20.0, P = 0.04). While the number of missed workdays was not significantly different for >1 hour of travel, the difference reached significance for those traveling >2 hours (24 days for >2 hours vs. 15 days for <2 hours, adjusted mean difference 10.5, 95% CI 2.1 to 18.8, P = 0.01). When assessing monthly missed days during the first 6 months (mean 14, SD 31, median 5, IQR 2,15), there were no significant differences either by rural/urban residence or by travel time.

The number of work days missed for each clinic appointment included choices for none, half, one, two, or more than 2 days. There was a significant increase in workdays missed for each visit for rural respondents (adjusted P\_trend = 0.02), those traveling >1 hour (adjusted P\_trend = 0.002), and those traveling >2 hours (adjusted P\_trend = 0.001).

Over one-third of respondents (36%) reported that at least one caregiver quit or changed jobs as a direct result of their child being diagnosed with cancer. When this was examined by residence and travel time, no significant differences emerged.

Another contributor to a caregiver's financial burden was the direct, out-of-pocket travel costs, including gasoline, airline or bus tickets, and hotel stays. The mean cost, per visit, was \$60.21 (SD \$100.6) for the entire cohort, but varied widely based on location of residence (rural = \$138 vs. urban = \$42, P < 0.001, travel time >1 hour = \$124 vs. <1 hour, travel time = \$19, P < 0.001, travel time >2 hours = \$168 vs. <2 hours = \$23, P < 0.001) (adjusted mean differences shown in Table III).

Participants were asked to rate their perception of the financial burden placed on them by their child's cancer using a standard visual analogue scale (0, no burden to 100, greatest possible burden) in Table III. The mean perceived financial burden for the entire cohort was 66 (SD 32, median 73, IQR 50,87). Multivariable models showed a significantly greater burden in rural respondents (adjusted mean difference 9.6, 95% CI 0.3 to 18.8, P = 0.04) and in those traveling >2 hours to the treatment center (adjusted mean difference 8.9, 95% CI 0.7 to 17.1, P = 0.03).

### Impact on Patient Education

Of caregivers of school-aged patients, 152 (60%) reported that their child was taken out of school during treatment. Of those continuing school, rural patients missed a more school days over the first 6 months from diagnosis than did urban patients (unadjusted means 90 vs. 58 days, respectively; adjusted mean difference 32.7, 95% CI 2.0 to 63.4, P = 0.04). While there was no significant difference in the average number of school days missed between those traveling >1 hour or <1 hour to the treatment center (unadjusted means 78 vs. 57 days, respectively; adjusted mean difference 19.4, 95% CI -11.1 to 48.0, P = 0.22), the difference was significant for those traveling >2 hours versus <2 hours (unadjusted means 87 vs. 57 days, respectively, P = 0.05; adjusted mean difference 32.8, 95% CI 3.6 to 62, P = 0.03) (Table III).

Even for patients staying in school, 28% of caregivers reported that their child was "not able to keep up with peers" and 10% reported repeating a grade, with rural patients being at an increased risk of repeating a grade (unadjusted percents 16% of rural patients

**TABLE II. Multivariable Poisson Regression Models for Dichotomous Outcomes: Use of Emergency Transportation and Burden in Childhood Cancer Patients' Families by Urban/Rural Residence and Travel Time**

Outcome	Rural vs. urban residence		Travel time to treatment center ≥1 hour vs. <1 hour		Travel time to treatment center ≥2 hours vs. <2 hours	
	Rate ratio	95% CI	Rate ratio	95% CI	Rate ratio	95% CI
Emergency transportation use <sup>a</sup>						
Any ambulance	1.6	0.9–2.8	1.6	1.1–2.6	1.6	1.0–2.5
Any emergency air transport	5.9	2.7–13.1	3.8	1.7–8.5	7.2	3.5–14.5
Burden measures <sup>a</sup>						
Relocated residence due to cancer	1.9	0.9–4.3	1.7	0.8–3.9	2.1	1.0–4.4
Caregiver quit work	1.0	0.6–1.7	0.9	0.6–1.3	0.9	0.6–1.5
Patient held back grade in school	3.0	1.1–8.3	0.9	0.2–3.1	1.5	0.5–4.7

<sup>a</sup>Each row represents different multivariable Poisson regression model adjusted for age at diagnosis, diagnosis, language, race, ethnicity, education and religion. Time since diagnosis was incorporated as the time variable in the Poisson models.

vs. 8% of urban patients; aRR 3.0, 95% CI 1.1–8.3,  $P=0.04$ ) (Table II).

### Impact of Diagnosis

The impact of patient diagnosis was also analyzed using the same outcomes of interest. Families of ALL patients were less likely to move because of cancer when compared to other diagnoses. Despite a small sample size, AML patients' families showed a trend toward being more likely to move (RR 2.0,  $P=0.08$ ) and were significantly more likely to move due to the cancer (RR 5.6,  $P=0.01$ ). In addition, caregivers of AML patients were more likely to quit work when compared to those with other diagnoses (RR 2.6,  $P<0.01$ ). There were no other significant differences seen between diagnosis groups (Supplementary Tables I and II).

### DISCUSSION

Our findings demonstrate a significant social and financial burden associated with caring for a child with cancer that, in many cases, is compounded by living remotely. While our institution serves a larger and less densely populated area than many pediatric oncology centers, most centers likely encounter similar challenges

in delivering care to geographically underserved populations. Of note is that a threshold of a travel time of over 2 hours was consistently associated with a greater impact on patients and families.

For urgent care, rural residence was not as sensitive a predictor of emergency ground transport utilization as was the travel time from the treatment center. However, the proportion of patients requiring air-transport was significantly increased in both rural and remote patients, with nearly one-quarter of rural patients requiring this costly intervention (estimated \$7,800–\$25,000 per flight) [19,20].

We found a surprisingly high percentage of caregivers who had moved residences because of their child's cancer. Given the considerable effort and financial costs associated with moving, the high proportion of families undertaking such a major life change highlights the considerable impact a cancer diagnosis has on the lives of patients' families. The trend towards having the move be more often "due to cancer" for those living further away suggests that the challenges encountered may be more pronounced for remote patients.

Our results indicate that having a child with cancer impacts caregivers' employment. Rural and remote caregivers clearly missed more workdays for each clinic visit than did proximal

**TABLE III. Multivariable<sup>a</sup> Linear Regression Models for Continuous Outcomes: Financial Burden and Missed Work and School for Childhood Cancer Patients' and Families, by Urban/Rural Residence and Travel Time**

Outcome	Rural vs. urban residence		Travel time to treatment center ≥1 hour vs. <1 hour		Travel time to treatment center ≥2 hours vs. <2 hours	
	Adjusted mean difference	95% CI	Adjusted mean difference	95% CI	Adjusted mean difference	95% CI
Perception of financial burden (visual analogue scale, 0–100)	9.6	0.3 to 18.8	3.3	–4.1 to 10.8	8.9	0.7 to 17.1
Travel expenses, \$	96	71 to 121	104	86, to 123	144	125 to 163
Missed work during first month of illness, days	10.6	1.1 to 20.1	6.3	–1.3 to 13.9	10.5	2.1 to 18.8
Missed work during first 6 months of illness, days	–2.1	–11.2 to 7.0	–1.0	–8.4 to 6.3	2.3	–5.8 to 10.3
Missed school during first 6 months of illness, days	32.7	2.0 to 63.4	19.4	–11.1 to 48.0	32.8	3.6 to 62

<sup>a</sup>Each row represents a different multivariable linear regression model adjusted for age at diagnosis, diagnosis, language, race, ethnicity, education and religion, as well as time since diagnosis.

patients. Interestingly, while remote caregivers missed more work than local caregivers during the first month after diagnosis, there was no significant difference in the amount of missed work between these groups when extended to include the first 6 months following the diagnosis. This finding suggests that many caregivers were potentially able to adjust to a new routine that accommodated their child's treatment schedule. However, sacrifices or compromises made to accomplish this were not captured by this survey.

Perhaps the most striking impact on employment we observed was that over one-third of the cohort quit or changed jobs as a direct result of their child having cancer, with no differences based on location of residence. As we do not have data on the caregivers' occupation, it is unclear if the amount of missed work or the risk of losing or changing jobs could be impacted by the nature or flexibility of a particular job type, which may differ between urban and rural populations. Also, it is likely that households with two working parents, a distinction not captured in this survey, would be impacted differently than those with a single wage earner.

We also found that the out-of-pocket expenses were substantially increased for remote families. As many patients require dozens of clinic visits over the course of therapy, a mean expense of \$138 per visit for rural patients can quickly add up to a significant financial burden. Accordingly, the perception of financial burden imposed by the child's cancer was significantly higher in rural families and those traveling greater than 2 hours. It is notable that the increased perception of financial burden in the remote patients exists despite an increased utilization of outside care in this population.

While patient diagnosis had no impact on most outcomes of interest, we found that AML had a greater impact on relocation and employment. This is likely due to the more intensive regimens and prolonged inpatient stays associated with AML therapy.

Our results are consistent with prior studies that have shown a significant financial burden for adult cancer patients and for caregivers of pediatric cancer patients [15,21–23]. However, to our knowledge, this study is among the first to document the impact of patients' residence on the financial burden of childhood cancer in the United States. As most pediatric cancer centers serve a larger geographic area than do adult centers, a significant proportion of patients travel great distances for cancer care. Some of the burden on rural families might be moderated by reducing the number of visits to the tertiary care center. In light of our findings, further research on the outcomes, costs, and benefits of strategies such as tele-health, home health services, involving local primary care physicians, and reducing frequency of follow-up visits after completion of therapy is warranted.

Our study's results also suggest a dramatic impact on schooling for the patients themselves. Over half elected to stop attending regular school while undergoing therapy. Of those continuing, rural patients missed more school days. While a travel time of >1 hour did not significantly impact the missed number of school days, traveling >2 hours did. The fact that 28% of patients had difficulty keeping up with their peers and 10% repeated a grade suggests that the demands of therapy significantly impact school performance, even for those continuing school. Our results also suggest the impact on schooling may be greater for rural patients as they more often repeated a grade. How childhood cancer's impact on patient education translates to more long-term financial implications remains unstudied.

As is the case with employment, school performance and attendance is multifactorial, and our analysis does not account for potential differences in academic rigor, supportive resources, or student populations among rural and urban schools. Much of the existing data regarding the impact of cancer therapy on education focuses on the effect of the treatment on learning ability, concentration, memory, and comprehension [24]. This underscores the need for more research on the impact of missed school and the development of interventions that maximize learning for children receiving complex cancer care. In particular, newer technologies, such as online video conferencing and Internet-based curriculums may be helpful to students, particularly those from remote populations.

Our study has several limitations. Sample size is a limiting factor, which precludes evaluating small subset of patients. A larger multi-institutional study would allow for a better understanding of the burden experienced by patients from extremely isolated communities. Related, this study only captures patients who were ultimately treated at a tertiary hospital, perhaps underestimating the challenges for patients without access to such an institution. Also, the study population is heterogeneous with respect to patient diagnoses, which could impact the results. However, our study aimed to explore outcomes that are common to all caregivers of pediatric oncology patients and all multivariable models included diagnosis to minimize the potential for confounding.

While our institution follows standard protocols common to most institutions, this sample is from a single institution and may not fully represent all patients. In addition, the survey was administered only to caregivers of patients who were alive at the time of measurement, which may not accurately portray the burden for those whose child succumbed to their disease.

Participants were not selected based on phase of treatment, which could influence responses, although many questions focused on the first 6 months from diagnosis to facilitate comparisons. While our regression models included time since diagnosis, future longitudinal work should assess these outcomes at specified time points to better understand how treatment phase may influence burden. While over 98% of surveys were completed by a parent, we did not assess whether it was the mother or father, which could have offered additional valuable information. Finally, with the exception of a significant proportion of Hispanic respondents, our survey did not capture the experiences of other minority groups.

In summary, our data show that having a child with cancer places a large burden on families, as is evidence by missed work, changes in employment, relocation of residences, travel expenses, and missed school. These burdens were greater for remote patients. Our study highlights issues unique to this underserved population and suggests the need for interventions that minimize the burden in such patients.

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