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ORIGINAL RESEARCH

Cost of survivorship care and adherence to screening—aligning the priorities of health care systems and survivors

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Abstract

Childhood cancer survivors (CCS) experience significant morbidity due to treatment- related late effects and benefit from late-effects surveillance. Adherence to screening recommendations is suboptimal. Survivorship care programs often struggle with resource limitations and may benefit from understanding institution-level financial outcomes associated with patient adherence to justify programmatic development and growth. The purpose of this study is to examine how CCS adherence to screening recommendations relates to the cost of care, insurance status, and institution-level financial outcomes. A retrospective chart review of 286 patients, followed in a structured survivorship program, assessed adherence to the Children's Oncology Group follow-up guidelines by comparing recommended versus performed screening procedures for each patient. Procedure cost estimates were based on insurance status. Institutional profit margins and profit opportunity loss were calculated. Bivariate statistics tested adherent versus nonadherent subgroup differences on cost variables. A generalized linear model predicted the likelihood of adherence based on cost of recommended procedures, controlling for age, gender, race, and insurance. Adherence to recommended surveillance procedures was 50.2%. Nonadherence was associated with higher costs of recommended screening procedures compared to the adherent group estimates ($2,469.84 vs. $1,211.44). Failure to perform the recommended tests resulted in no difference in reimbursement to the health system between groups ($1,249.63 vs. $1,211.08). For the nonadherent group, this represented $1,055.13 in "lost profit opportunity" per visit for patients, which totaled $311,850 in lost profit opportunity due to nonadherence in this subgroup. In the final model, nonadherence was related to higher cost of recommended procedures ($p < .0001$), older age at visit ($p = .04$), Black race ($p = .02$), and government-sponsored insurance ($p = .03$). Understanding institutional financial outcomes related to patient adherence may help inform survivorship care programs and resource allocation. Potential financial burden to patients associated with complex care recommendations is also warranted.

Keywords

Childhood cancer, Adherence, Survivorship care, Financial burden, Cost of care

Implications

Practice: Efforts to improve adherence to survivorship care guidelines among childhood cancer survivors should consider addressing the financial burden associated with costs of care, such as assisting patients in understanding, being prepared for, and managing health care costs.

Policy: Understanding the ways in which patient adherence is related to the cost of care and financial burden to patients, and financial impact on healthcare institutions, may be important to building institutional support for survivorship care programs and resources for optimizing adherence rates.

Research: Further evaluation of the ways in which financial factors have an impact at the patient level and at the institution level may be important to building programs that will support optimal patient adherence and survivorship care.

INTRODUCTION

Over 80% of children diagnosed with cancer are cured of their disease resulting in more than 350,000 childhood cancer survivors (CCS) living in the USA [1, 2]. As a consequence of exposure to curative treatment modalities, such as chemotherapy, radiation, and surgery, CCS experience high rates of morbidity and mortality due to secondary health problems [3]. As many as 95% of CCS will develop a chronic disease, with over 80% developing a severe, disabling, or life-threatening condition, due to treatment effects [4, 5]. Despite the Children's Oncology Group (COG) development of exposure- and risk-based screening guidelines for the long-term follow-up of CCS [6], adherence to posttreatment screening recommendations remains suboptimal even among high-risk survivors [7]. Less than a third of CCS report receiving follow-up care that is based on their cancer history [8]. Understanding how patient adherence relates to hospital-level finances may be critical to establishing survivorship care programs and optimizing the long-term follow-up and health outcomes of this at-risk group.

Survivorship programs that promote evidence-based surveillance of late effects lead to early detection, referral, and treatment. Program activities include providing patients with up-to-date information about late effect, educating about

care recommendations, and developing and implementing personalized follow-up plans [9]. Despite the well-recognized need of long-term follow-up care for CCS, programs are often limited by inadequate resources and understaffing. Decisions within health care systems about resource allocation are influenced by revenue generation. Survivorship care can be labor and time intensive and, in the USA, reimbursement for survivorship visits and financial remuneration is limited compared to other service lines. While some services are revenue generating (e.g., surveillance testing), many are not (e.g., care coordination), and the economic case for program benefit is considered tenuous [10, 11]. Many cancer-treating institutions have limited capacity to support the long-term follow-up of CCS patients [12]. Financial constraints and lack of administrative support are cited as barriers to the adoption and growth of survivorship programs [13]. Developing and sustaining such programs typically requires financial backing and support from the institution for services, staff, and space allocation [14]. It is argued that the cost evaluation of survivorship programs is essential to justify program value and to garner administrative buy-in and institutional support, but this has been limited to date [15, 16].

We previously described rates of adherence to survivorship care recommendations, based on COG guidelines, within a structured, long-term follow-up clinic for CCS [11–13, 17]. Given the importance of surveillance and the vital role of survivorship programs in providing cohesive long-term care for survivors, these secondary analyses from the same data set compared institution-level financial outcomes between adherent and nonadherent patient subgroups regarding reimbursement, profit margin, and profit opportunity loss. We also examined how the cost of care and insurance status related to adherence to survivorship care recommendations, controlling for relevant covariates. To our knowledge, this is the first study to examine differences in institutional financial outcomes based on patient adherence to COG guidelines, including health system-level profit margins. Results from this analysis may inform programmatic decision-making within health care systems and drive institution- or policy-level changes to support clinical efforts to improve patient adherence and the long-term survivorship care of CCS.

METHODS

This study was conducted within the CCS long-term survivorship program within the Division of Pediatric Hematology/Oncology and Stem Cell Transplant at Cohen Children's Medical Center, Northwell Health. All study procedures were approved by the Institutional Review Board and the Cancer Services Scientific Review Committee of the Northwell Health System.

This was a secondary analysis of data collected from a retrospective chart review of 286 patients followed in a structured, long-term follow-up program for CCS housed within a large, integrated medical center in the New York area. Within this program, patients are seen in a medical office, off-campus from the main children and adult hospitals. Clinic visits include a review of late-effect risks and recommended screening procedures with a physician or nurse practitioner that specializes in survivorship care, a laboratory evaluation (at an on-site phlebotomy center), meeting with a nurse coordinator to facilitate the scheduling and performance of screening procedures (e.g., echocardiography and pulmonary function testing breast magnetic resonance imaging [MRI]), and meeting with a social worker to address psychosocial concerns. Screening recommendations and referrals are generally based on the COG guidelines [18].

Study population

All CCS followed in the survivorship program were eligible for the study. Patients are eligible to be seen in the program once they are 3 years from the completion of their cancer therapy and disease free. The program provides lifelong follow-up care.

Data collection

This study included secondary analysis of data obtained in a retrospective review of the medical records of all CCS seen at least once in the survivorship program between January 1, 2010 and December 31, 2012. Data included standard sociodemographic information, cancer history and treatment exposure, and insurance coverage (categorized into three groups: commercial insurance, an "underinsured" group representing Medicaid or no insurance, and unknown). Preliminary analyses did not identify differences between subgroups with Medicaid, no insurance (self-pay), and unknown in rates of adherence (p = .48). For each patient, survivorship care documentation included recommended and completed procedures.

Based on COG guidelines, procedures included the following screening tests: echocardiography, electrocardiograms, pulmonary function tests, dual-energy x-ray absorptiometry scans (DXA) scans, breast MRI, breast mammography, hearing tests, fasting blood work, nonfasting blood work, and urinalysis. In addition, we included breast ultrasound, as this is often a patient-preferred screening modality to mammography, and thyroid ultrasound, as this was routinely recommended in our survivorship practice despite it not being specifically recommended by the COG guidelines. Financial data about procedure costs and reimbursement to the health system were obtained to evaluate the projected and actual costs of procedures (recommended and performed) and reimbursement to the health system based on insurance status.

Variable definitions

Adherence

Adherence to a given recommendation was defined as completing the screening procedure within 12 months of the recommendation, given that most survivorship visits and screening recommendations occur on an annual basis. If patients had survivorship visits more frequent than annually, adherence was defined as having the screening test performed prior to the next visit for fasting blood work, nonfasting blood work, and urinalysis. Adherence is reported at the patient level (two groups; adherent vs. nonadherent) and at the visit level by procedure. At the patient level, nonadherence was defined as the failure to complete any one recommended procedure such that patients that completed some but not all recommendations were still classified as nonadherent. Patients that were recommended to complete more than one procedure within the study time frame were defined as adherent only if they completed all recommendations. Adherence at the visit level was also reported as it was theorized that patients' adherence may change over time or vary depending on the type of procedure recommended, and this allowed us to model factors with varying effects on adherence across visits.

Survivorship care costs

Financial records of the Northwell Health System during the 3 year time frame were reviewed to quantify the cost and reimbursement for screening tests provided that were directly related to survivorship care within the survivorship program. Four variables were calculated. Two variables described procedures that were recommended for each patient: the projected cost of recommended procedures (i.e., fee estimate to the health system) and the reimbursement amount for recommended procedures (i.e., projected value the health system would receive). Two additional variables described procedures that were completed by each patient: the cost of performed procedures (i.e., fee estimate for performed procedures to the health system) and the reimbursement amount for performed procedures (i.e., the value the health system receives for each test performed). Variables estimating the cost of and reimbursement for recommended procedures followed COG guidelines for surveillance testing for each patient, irrespective of whether patients completed the procedure. Variables representing the cost of and reimbursement amount for performed procedures depended on patient adherence and whether the procedure was actually performed.

Following this, variables were calculated to estimate the institutional profit when patients were adherent and, conversely, the "lost profit opportunity" when patients were nonadherent. The profit of performed procedures was calculated by subtracting the cost of performed procedures from the reimbursement of performed procedures. The health system's lost profit opportunity was defined by first calculating what the profit would have been if all recommended procedures were completed for each patient and subtracting the profit that was actually made from the procedures that were done. Thus, this opportunity loss represented the profit loss associated with nonadherence.

Data analysis

Descriptive statistics were used to describe the study population, adherence rates, costs of recommended/performed procedures, and reimbursement. Bivariate analyses identified differences between adherent and nonadherent patient subgroups based on sociodemographic characteristics and cost variables, including group differences in health system-level profit and profit opportunity loss. Adherence was analyzed separately for all recommended surveillance tests with the exception of breast MRI, breast ultrasound, and mammography due to the small number of these tests recommended in this study group. Analyses used independent samples *t*-tests and chi-square to test bivariate associations. Understanding that our preliminary analyses testing bivariate associations would be unable to account for nonindependence of data, a generalized linear model (GLM) was used to predict likelihood of adherence using a generalized estimating equation (GEE) to derive parameter estimates as detailed below. In the GEE models, we used binary distribution and logit link function for logistic regression. We used TYPE=EXCH option to specify an exchangeable working correlation structure; the CORRW option to request the working correlation matrix and LOGOR=fullclust for log odds ratio (OR); and quasi-information criterion was used for goodness of fit.

To evaluate associations between survivorship care cost and adherence, while controlling for relevant covariates, such as age and cancer diagnosis, multivariate models were specified with adherence as the outcome variable. A GLM was used to predict the likelihood of adherence using a GEE to derive parameter estimates as this type of model is appropriate when there is nonindependence within the covariance structure such that correlated data due to patients having multiple visits were accounted for. Adherence measured at the visit level was used as the outcome variable in the final multivariable model in order to evaluate factors related to adherence that vary within patients over time (e.g., age). The visit-level cost of recommended procedures was included as a predictor of adherence as a two-group categorical variable based on a median split ($400). All models controlled for age at visit, gender, race/ethnicity, and insurance status.

RESULTS

The charts of 286 patients seen in the survivorship clinic for long-term follow-up care after childhood cancer were reviewed, representing a total of 542 visits during the 3 year period. Study sample characteristics, diagnosis, and treatment data are in Table 1. The average age of diagnosis was 7.9 years; 50.4% were male and 19.6% were of Medicaid or self-pay insurance status. The overall rate of adherence to recommended procedures was 50.18% (272 out of 542 recommended procedures), with 74.2% of patients adherent to all recommended procedures (212 out of 286 patients).

Patient adherence to screening recommendations varied based on several sociodemographic and medical factors (Table 2). In a separate bivariate analysis, classification into the nonadherent group was associated with older age at visit (Means = 18.0 vs. 16.5 years old), identifying as Black (vs. Other), and being underinsured (i.e., Medicaid or no insurance; vs. commercial, private, or unknown insurance). Age at diagnosis, gender, and history of radiation therapy (RT), anthracycline, and transplant was not related to recommendation adherence. Rates of adherence were similar across recommended surveillance tests ($p > .05$).

Mean costs of recommended and performed procedures, including health system-level profit and profit opportunity loss, and differences between adherent and nonadherent groups are reported in Table 3. The average cost of recommended procedures for the adherent group was $1,211.44 (standard deviation [SD] = $1,272.27), whereas the projected cost of recommended procedures for the nonadherent group was significantly higher at $2,469.84 (SD = $2,100.68). Thus, the nonadherent group was characterized by greater potential costs at the institution level if they were to undergo all recommended procedures compared to the adherent group. There was no difference between adherent and nonadherent groups in the reimbursement for procedures performed, which averaged just over $1,200 for each group (i.e., $1,211.08 in the adherent group and $1,249.63 in the nonadherent group).

Further comparison of the adherent and nonadherent groups evaluated the degree of profit and profit opportunity loss at the institution level. It was estimated that, per visit, the health system received an average of $850.52 (SD = $1,135.23) in profit per visit for patients in the adherent group. Conversely, the nonadherent group's recommended care included higher cost procedures with a greater potential profit margin for the health system. It was estimated that the health system would have received an average of $1,932.81 (SD = $1,924.22) in profit per visit *if* these patients had completed all recommended procedures. However, because they were not fully adherent, the profit per visit for the nonadherent group averaged $877.68 (SD = $1,411.68) similar to that of the adherent group. Thus, the average lost profit opportunity per visit for patients within the nonadherent group was $1,055.13 (SD = $1,545.74; i.e., $1,932.81 – $877.68). The total lost profit to the health system

Table 1 | Patient characteristics ($N = 286$)

Patient characteristics	Mean (range)	*n* (%)
Average age at diagnosis	7.9 (0–23)	
Gender		
Male		142 (49.6%)
Female		144 (50.4%)
Race		
Asian		20 (7.0%)
Black		29 (10.1%)
Hispanic		30 (10.5%)
White		112 (39.2%)
Other		10 (3.5%)
Unknown		85 (29.7%)
Insurance		
Commercial		177 (61.9%)
Medicaid or self-pay		56 (19.6%)
N/A		53 (18.5%)
Cancer diagnosis[a]		
Leukemia[b]		124 (43.4%)
Lymphoma		78 (27.3%)
Bone and soft tissue tumors		20 (7.0%)
Embryonal tumors		50 (17.5%)
Others		14 (4.9%)
Treatment		
Chemotherapy (CT) only		165 (57.7%)
Radiation therapy (RT) only		2 (0.7%)
Both CT and RT		117 (40.9%)
Neither CT nor RT		2 (0.7%)
Total visits at survivorship clinic		
1		119 (41.6%)
2		92 (32.2%)
3		64 (22.4%)
4		8 (2.8%)
5		3 (1.0%)
Type of treatment		
Alkylator		186 (65.0%)
Carboplatin		18 (6.3%)
Cisplatin		33 (11.5%)
Etoposides		104 (36.4%)
Antimetabolites		158 (55%)
Dactinomycin		23 (8.0%)
Vincaalkyloids		230 (80.4%)
Anthracycline		216 (75.5%)
Corticosteroids		185 (64.7%)
Transplant		16 (5.6%)

[a]Brain tumor survivors were managed in a separate brain tumor program.
[b]Includes B acute lymphoblastic leukemia (B-ALL), T acute lymphoblastic leukemia (T-ALL), acute myelogenous leukemia (AML), and chronic myelogenous leukemia (CML).

Table 2 | Differences between adherent and nonadherent patient groups: sociodemographic and medical factors

	Adherent group (n = 272 visits)	Nonadherent group (n = 270 visits)	p value
	M (SD) or n (%)	M (SD) or n (%)	
Patient characteristics			
Average age at visit (years)	16.5 (6.9)	18.0 (7.3)	.0106
Gender			.2295
Male	145 (52.7%)	130 (47.3%)	
Female	127 (47.6%)	140 (52.4%)	
Race[a]			.0825
Asian	19 (46.3%)	22 (53.7%)	
Black	19 (35.9%)	34 (64.1%)	(.028)[b]
Hispanic	28 (49.1%)	29(50.9%)	
White	124 (55.6%)	99 (44.4%)	
Other	13 (65.0%)	7 (35.0%)	
Unknown	69 (46.6%)	79 (53.4%)	
Insurance[a]			.0105
Commercial	185 (55.1%)	151 (44.9%)	
Medicaid or self-pay	43 (39.5%)	66 (60.5%)	
N/A	44 (45.4%)	53 (54.6%)	
Cancer diagnosis			.0451
ALL	128 (53.6%)	111 (46.4%)	
Lymphoma	57 (39.9%)	86 (60.1%)	
Bone and soft tissue tumors	19 (46.3%)	22 (53.7%)	
Embryonal tumors	57 (57.0%)	43 (43.0%)	
Others	11 (57.9%)	8 (42.1%)	
Treatment			.9989
Chemotherapy (CT) only	159 (50.5%)	156 (49.5%)	
Radiation therapy (RT) only	1 (50.0%)	1 (50.0%)	
Both CT and RT	111 (49.8%)	112 (50.2%)	
Neither CT nor RT	2 (50%)	1 (50%)	
Total visits 1			<.001
1	37 (31.1%)	82 (68.9%)	
2	80 (43.5%)	104 (56.5%)	
3	125 (65.1%)	67 (34.9%)	
4	24 (75.0%)	8 (25.0%)	
5	6 (40.0%)	9 (60.0%)	
Type of treatment			.5626
Alkylator			
Yes	179 (49.3%)	184 (50.7%)	
No	93 (52.0%)	86 (48.0%)	
Carboplatin			.2769
Yes	25 (58.1%)	18 (41.9%)	
No	247 (49.5%)	252 (50.5%)	
Cisplatin			.9736
Yes	35 (50.0%)	35 (50.0%)	
No	237 (50.2%)	235 (49.8%)	
Etoposides			.6733
Yes	99 (49.0%)	103 (51.0%)	
No	173 (50.9%)	167 (49.1%)	
Antimetabolites			.5516
Yes	155 (51.3%)	147 (48.7%)	
No	117 (48.7%)	123 (51.3%)	

Table 2 | Continued

	Adherent group (n = 272 visits)	Nonadherent group (n = 270 visits)	p value
	M (SD) or n (%)	M (SD) or n (%)	
Dactinomycin			.1779
Yes	28 (59.6%)	19 (40.4%)	
No	244 (49.3%)	251(50.7%)	
Vincaalkyloids			.4021
Yes	217 (49.3%)	223 (50.7%)	
No	55 (53.9%)	47 (46.1%)	
Anthracy			.4343
Yes	208 (49.3%)	214 (50.7%)	
No	64 (53.3%)	56 (46.7%)	
Corticosteroids			.4543
Yes	173 (49.0%)	180 (51.0%)	
No	99 (52.7%)	90 (47.6%)	
Transplant			.3115
Yes	21 (58.3%)	15 (41.7%)	
No	251 (49.6%)	255 (50.4%)	
Annual visit			.0140
Yes	208 (47.6%)	229 (52.4%)	
No	64 (60.95%)	41 (39.05%)	

ALL acute lymphoblastic leukemia; *SD* standard deviation.
[a]High rates of missing data were observed for Race and Insurance, which was adjusted for by including the categories "Unknown" and "N/A" into group difference tests, respectively.
[b]p value for Black versus all other races/categories

Table 3 | Differences between adherent and nonadherent patient groups: economic factors for the health care system[a]

	Adherent group (n = 272 visits)	Nonadherent group (n = 270 visits)	p value
Economic factors[a]			
Expected reimbursement to the health system based on procedures recommended ($)	1,211.4	2,469.8	<.0001
Actual reimbursement to the health system based on procedures performed ($)	1,211.4	1,249.6	.7529
Expected cost to the health system procedures recommended ($)	362.0	537.0	<.0001
Actual cost to the health system of procedures performed ($)	362.0	372.0	.4229
Expected profit based on recommended procedures[b] ($)	849.4	1,932.8	<.0001
Actual profit for performed procedures[c] ($)	849.4	877.6	.8053
Lost opportunity[d] ($)	0	1,055.1	<.0001

[a]Reported in U.S. dollars ($).
[b]Expected profit for recommended procedures = expected reimbursement of recommended procedures − expected cost of recommended procedures.
[c]Actual profit for performed procedures = actual reimbursement of performed procedures − actual cost of performed procedures.
[d]Lost opportunity = expected profit for recommended procedures − actual profit for performed procedures.

due to nonadherence was $311,850 for these data set representing patient visits within this 3 year time period.

Multivariate model predicting adherence

Results of the multivariate analysis are shown in Table 4 (GEE model fit criteria, quasi-information criterion = 691.80). The cost of recommended procedures was found to be a highly significant predictor of adherence (p < .001). Patients with recommended procedures that exceeded $400 (median cost) were less likely to be adherent than those with recommended procedures that cost less than $400 (OR = 0.32; 95% confidence interval [CI]: 0.215 to 0.464). Evaluation of covariates also indicated that all factors associated with adherence in bivariate analysis remained significant in this overall model. Older age at visit (OR = 0.97; 95% CI: 0.943 to 0.998), Black racial/ethnic identity (OR = 0.47; 95% CI: 0.232 to 0.902), and underserved insurance status (OR = 0.59; 95% CI: 0.361 to 0.955) related to greater likelihood of nonadherence. Cancer

Table 4 | Multivariate model predicting adherent versus nonadherent subgroup membership ($N = 542$)

Patient Characteristics	Estimate coefficient	*p* value	Odds ratio	95% confidence interval
Black race (vs. all other races)[a]	−0.7827	.0240	0.4572	(0.2317, 0.9019)
Underserved insurance (vs. commercial insurance)[a]	−0.5325	.0319	0.5872	(0.3610, 0.9550)
Age at visit	−0.0305	.0360	0.9699	(0.9426, 0.9980)
Male gender (vs. female gender)	0.1250	.5450	1.1332	(0.7559, 1.6988)
Cost of recommended procedures ≥$400 (vs. ≤$400)[b]	−1.1517	<.0001	0.3161	(0.2154, 0.4639)

[a]High rates of missing data were observed for race and insurance, which was adjusted for in the model by including missing data categories in the categorical variables.
[b]$400 was the median value of the cost of recommended procedures at the visit level.

diagnosis was not included in the multivariate model due to multicollinearity with age and cost of recommended procedures.

DISCUSSION

Survivors of childhood cancer are at increased risk for chronic health conditions and secondary malignancies that require long-term follow-up surveillance. Adherence to exposure- and risk-based screening guidelines improves detection of late effects and can be cost effective [5, 15, 19]. We found that nonadherence to screening procedures among CCS resulted in lowered reimbursement amounts to the health system, which we termed "lost profit opportunity." Patients were less likely to be adherent to higher cost procedures. Other factors relating to nonadherence included older age, identifying as Black, and being underinsured. Improving CCS adherence to screening recommendations is an important undertaking. Ultimately, it will require broad efforts both at the patient level, by reducing barriers and targeting those patients most at risk for nonadherence, as well as at the system level through the allocation of resources and institutional support for survivorship care programs, such as patient navigators and financial counselors.

Impact to the health system

Caring for CCS requires a coordinated, multidisciplinary approach and involves screening that utilizes multiple modalities, including blood tests, radiology tests, and evaluation by subspecialists. Each of these health care utilization encounters provides an opportunity for financial profit to the health care institution. When CCS are nonadherent to screening recommendations, this results in fewer health care encounters and, therefore, represents a "lost profit opportunity" for the health system. In this study, we found that the total lost profit to the health system due to nonadherence was $311,850 for 286 CCS during the 3 year time frame of this study (2010–2012). This translates to approximately $348,700 in today's dollars, adjusting for inflation from 2012 to 2019.[20] It is notable that survivorship care programs have been characterized as limited in their revenue-generating opportunity. However, findings suggest opportunity for some financial benefit to the hospital. Clearly, hospitals have both clinical and financial incentives to improve adherence and profit margin. Despite established care guidelines, clinicians treating CCS may be in positions of needing to justify the value of survivorship program. This is consistent with research citing program development and resource factors (e.g., hiring staff and finding space) as the most commonly cited need among survivorship care teams [14]. Greater investment in survivorship programs that increase clinic resources, allowing for supportive care efforts to address patient-level access barriers, may translate to improved patient adherence and increased profit.

The association between lost profit opportunity and adherence amongst CCS provides an opportunity for mutual benefit. A modest investment of resources on the part of the health system to improve adherence amongst CCS can provide a positive return on investment. It is, thus, important to clearly identify the specific barriers to adherence among CCS, and we have begun a follow-up study of our survivorship population to attempt to do so. In the interim, it is reasonable to speculate that improving access to screening tests (e.g., through improved patient navigation, care coordination that provides facilitated scheduling, and ease of transportation to the testing site) would likely improve adherence. Communication between survivorship teams and health system administration surrounding the barriers to adherence may be able to relieve some of those barriers with a minimal investment of time and resources, thereby improving the health and quality of life in the CCS and providing a return on investment for the health system.

Notably, access to care and patient adherence exists within the broader social ecology of health and health care in the USA, which includes individual, family, and societal factors, payer systems, policy, and government structures [21]. In this study, factors specific to our institution (e.g., scheduling problems) or location (e.g., commuting challenges) may have impacted patient adherence. Among CCS, adherence to surveillance recommendations is influenced by geographic location and the availability of providers and cancer care settings [22]. Data were also collected prior to many of the provisions mandated by the Affordable Care Act (ACA). Although research shows that the ACA has

substantially decreased the number of uninsured individuals in the USA, changes vary by state and the affordability of health care, and financial toxicity effects of cancer treatment, in particular, are still a concern [23–26]. As changes continue to occur with respect to health care delivery and financing, further research is needed to understand the impact on access to care among CCS [27]. Analysis of the cost effectiveness of COG guidelines within the context of health care system changes, including downstream health care costs and financial burden to patients, may be important for refining recommendations, developing targeted approaches to support patient adherence, and guiding the long-term follow-up care of CCS [15, 28, 29]. Representing the pharmaceutical industry, insurance providers, oncologists, and patient advocates, Zafar et al. present potential solutions for the rising cost of cancer care, targeting multiple levels, such as increasing price awareness among providers, the inclusion of cost in treatment discussions with patients, and policy interventions (e.g., value-based contracting), citing that all stakeholders must be involved to ensure high-value care [30]. Although we showed modest profit margins and profit opportunity related to adherence within our survivorship care program, addressing these broader challenges is critical as institutions make decisions about resource allocation and investment in clinical services.

Impact to the patient

We previously identified suboptimal rates of adherence to personalized, risk-based screening recommendations for CCS [17]. In this follow-up study, we found several patient-level factors correlated with nonadherence, including older age, identifying as Black, and government-sponsored insurance. Other patient factors predicting nonadherence among CCS include knowledge of treatment exposures, perception of symptoms and risk, motivation, affect, and provider influences [31–33].

We also found the cost of care at the hospital level distinguished survivors' likelihood of being adherent even when controlling for other sociodemographic factors and insurance type. There appeared to be a financial "threshold" of ~$1,200 that characterized costs in both the adherent and nonadherent groups, above which the nonadherent group was noncompliant with recommended care. Recommended care that includes more procedures or higher cost procedures likely represents a greater burden to patients, such as the time, effort, and/or finances required. Trends over the last several decades have shown increased cost sharing between patients and payers, including increasing deductibles, increasing premiums, coinsurance, greater copayments, and reduced affordability [30]. Combined with increasing costs of medication and treatment, it is well documented that shifts in payment structures have yielded greater financial stress for cancer survivors [34]. Among CCS, higher out-of-pocket medical costs and the experience of personal financial problems are associated with delaying or foregoing care, including skipping surveillance tests, treatment, and follow-up [30, 31]. Kent et al. reported that self-reported financial problems led to an almost 200% increase in the proportion of survivors who skipped medical care compared to those without financial problems (13.8% vs. 5.0%, respectively) [35].

This study is limited as the cost of care to patients was not quantified. However, there is often a financial burden for patients to attend clinic visits and adhering to screening guidelines. Financial cost of healthcare is a well-known factor contributing to nonadherence across patient populations, including cancer survivors [36]. Compared to noncancer peers and siblings, CCS also have lower socioeconomic attainment, including lower education, higher rates of under/unemployment, and lower income [37], which is associated with less use of follow-up medical care [38]. More than half of adult survivors of childhood cancer report concern about being unable to pay for health care and medication expenses and 33% report an inability to obtain care due to finances [39]. Within the Childhood Cancer Survivor Study (CCSS), 25% of survivors had skipped a medical test, treatment, or follow-up care in the past year because of cost; 28% postponed preventative care; and 16% did not fill a prescription because of cost [40]. Forgoing care and nonadherence to surveillance recommendations undermine evidence-based care for health risk management and jeopardize CCS's long-term quality of life [35, 41, 42].

Further work is needed to better understand patient- and system-level factors that impact CCS's adherence behaviors and to build supportive care strategies to improve adherence rates. This may be particularly important for high-risk patient subgroups.[43] At minimum, efforts must be made to ensure that patients have an accurate understanding of health risks and surveillance recommendations, as misperceptions of susceptibility to late effects have been reported at high rates [44]. Patients may also benefit from resources to help them understand cancer-related finance issues, such as health care bills and insurance coverage, to help with financial planning, and to overcome barriers to accessing care (e.g., low-cost travel arrangements) [45, 46].

Several strategies may be implemented to identify patients at risk for nonadherence and provide meaningful support. Brief screening tools or question prompts for providers may feasibly be incorporated into routine care to identify and triage patients in need of health system-, community-, or web-based support resources [39, 47]. Educational and skills-building interventions are needed to help promote optimal health management and planning among

patients or to guide patients through complex decisions, such as choosing health insurance plans that meet their needs. Park et al. [48] conducted in-depth interviews with CCS about their perceptions and knowledge of the ACA and found pervasive knowledge deficits and concerns about future health care costs among both insured and uninsured survivors. As CCS are at risk for financial barriers to care, patient-centered financial education courses and provider- or staff-level training ("financial navigators") may provide significant benefit in this respect [49–51]. Resources developed by patient organizations exist to educate and support survivors around cancer-related financial toxicity, including health insurance selection (e.g., Cancer Financial Assistance Coalition). Shankaran et al. in partnership with the Consumer Education and Training Services (CENTS) and Patient Advocate Foundation (PAF) developed a financial education course for cancer patients, aimed to improve knowledge of treatment costs, provide financial counseling, and assist in managing out-of-pocket expenses [51, 52]. A recent systematic review reported that only two of five randomized controlled trials aiming to improve CCS's adherence to surveillance guidelines were effective; both involved telephone outreach, suggesting the importance of clinic resources and adequate personnel time to make such services feasible within survivorship care programs [53]. Further research is needed to develop evidence-based resources and to validate those that already exist to determine their efficacy in helping patients understand and manage their finances and to support financial decision-making.

Limitations

Our study is limited by the relatively small cohort, in which patients were actively engaged in survivorship care. The relatively short follow-up time period limited our ability to evaluate the impact of patient adherence and surveillance on long-term costs or health outcomes for patients. We also observed that the proportion of adherent patients increased with a greater number of visits, whereas the proportion of nonadherent patients decreased. This may have been due to: (a) those who are recommended to be seen more frequently are typically much closer in time from the end of therapy and may have a heightened concern about their health and/or (b) those who are seen more often receive more counseling, are perhaps more educated and vested in their care, and are more connected to the survivorship care team. We were unable to obtain data from survivors who did not attend a clinic visit at least once or who dropped out and findings may be biased toward those willing and financially able to attend the clinic, representing a greater likelihood of adherence. Childhood cancer survivors, particularly those in the young adult age range, are a transient group, and tracking patients over time can be difficult. As hospitals increasingly adopt digital health strategies for connecting with patients, systems that support transportable survivorship care and data collection is critical for clinical and research efforts.

As there are many models of survivorship care delivery, we may not be able to extrapolate our adherence costs to other programs. Our patient cohort was also primarily White with commercial insurance and there was missing data on patient-level factors due to gaps in data in the electronic medical record. Missing data were a limitation, particularly, for race and insurance variables. However, this was accounted for in the analyses by including missingness into the models (i.e., "Unknown" and "N/A" categories for race and insurance, respectively) such that parameter estimates were generated controlling for missing data. Nevertheless, this presents a limitation to our findings. We were also unable to directly measure the financial impact of survivorship care on the patient (e.g., copays and out-of-pocket expenses) as the actual cost to the survivor is a function of a patient's insurance coverage. The insurance data we incorporated into our analyses were based on estimates and do not reflect individual patients' cost burden. Data also do not include hospital-level costs associated with comorbidities, which further limits our findings as this patient population is at increased risk for comorbid health problems. We based our financial data on health system-specific figures, assuming all CCS had their surveillance procedures done within our health system. As procedure costs vary among practice settings, we may not be able to extrapolate our cost data to other institutions.

CONCLUSION

In conclusion, we believe that dedicated survivorship programs are important in the care of CCS. Resourcing those programs is challenging, as they are often loss leaders in that they do not generate revenue for the health system through their own billing. Nonetheless, CCS undergo multiple screening tests, all of which are revenue generating and would likely not be performed within the health system if not for the presence of the survivorship program. As such, while it can be uncomfortable, it is important to highlight the fiscal contributions of survivorship programs to the health system to justify resourcing them most appropriately. To that end, we have identified the cost of care as a factor associated with adherence in CCS, and that adherence is, in turn, associated with a better financial outcome for the health system. This relationship suggests that finding ways to improve adherence to care among CCS would be mutually beneficial to the patient and the health system. Some possibilities include the establishment of financial coordinators who can improve access for CCS to materials and education surrounding insurance options and managing out-of-pocket expenses, improving the coordination of

screening tests to reduce days of missed work and travel expenses, and supporting research in understanding the barriers CCS face in adhering to screening recommendations.

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Compliance with Ethical Standards

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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. For this type of study, formal consent is not required. All study procedures were approved by the Institutional Review Board and the Cancer Services Scientific Review Committee of the Northwell Health System. This article does not contain any studies with animals performed by any of the authors.

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