

Cancer Prevention and Screening Practices of Siblings of Childhood Cancer Survivors: A Report from the Childhood Cancer Survivor Study

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Abstract

Background: To compare the skin and breast/cervical cancer prevention/screening practices of adult siblings of childhood cancer survivors with controls and to identify modifying factors for these practices.

Methods: Cross-sectional, self-report data from 2,588 adult siblings of 5+ year survivors of childhood cancer were analyzed to assess cancer prevention/screening practices. Two age, sex, and race/ethnicity-matched samples (N = 5,915 and N = 37,789) of the Behavioral Risk Factor Surveillance System participants served as the comparison populations. Sociodemographic and cancer-related data were explored as modifying factors for sibling cancer prevention/screening practices through multivariable logistic regression.

Results: Compared with controls, siblings were more likely to practice skin cancer prevention behaviors: use of protective clothing [OR, 2.85; 95% confidence interval (CI), 2.39–3.39], use of shade (OR, 2.11; 95% CI, 1.88–2.36), use of sunscreen (OR, 1.27; 95% CI, 1.14–1.40), and wearing a hat (OR, 1.77; 95% CI, 1.58–1.98). No differences were noted for breast/cervical cancer screening including mammography and Pap testing. Having less than a high school education and lack of health insurance were associated with diminished cancer prevention/screening behaviors. Survivor diagnosis, treatment intensity, adverse health, chronic health conditions, and second cancers were not associated with sibling cancer prevention/screening behaviors.

Conclusions: Siblings of cancer survivors report greater skin cancer prevention practices when compared with controls; however, no differences were noted for breast/cervical cancer screening practices. Access to care and lack of education may be associated with decreased cancer prevention/screening behaviors. Interventions are needed to address these barriers.

Impact: Research should be directed at understanding the impact of the cancer experience on sibling health behaviors. *Cancer Epidemiol Biomarkers Prev*; 21(7); 1078–88. ©2012 AACR.

Introduction

Having a sibling with childhood cancer can influence psychological and behavioral outcomes in adulthood (1).

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Estimates suggest that in 2005 there were more than 325,000 survivors of childhood cancer in the United States (2). The National Cancer Institute, Office of Cancer Survivorship states that "family members, friends, and caregivers are part of the survivorship experience" (3). With the excellent survival rate for childhood cancers and the average U.S. family size of 2 children, (4) there is also a large and growing population of siblings of childhood cancer survivors that continue to be impacted by the cancer experience long-term.

Research with siblings of children on active cancer therapy shows that siblings experience stress and losses such as loss of parental availability and unmet needs (5–12). Associated with this stress as well as losses and unmet needs are minimization and underreporting of the health care needs of siblings (12). The long-term impact of this minimization and underreporting, and its effect on cancer prevention/screening in siblings has not been studied. Models of health behavior suggest that this minimization and underreporting of health care needs as a child may predict decreased adult screening and prevention

practices (13, 14). Research shows that siblings of survivors are also at higher risk for developing cancer (15, 16) and adverse health behaviors such as alcohol use (17, 18) that are associated with increased risk for the development of specific cancers.

Childhood experiences with significant stress have the ability to undermine adaptation, the development of coping skills, and social networks (19–21). Furthermore, stressful childhood experiences are associated with the development of unhealthy lifestyles. In the context of the childhood cancer experience, research shows that siblings experience significant stress resulting in impaired adaptation, coping, and social relationships. Adult siblings of childhood cancer survivors also show unhealthy lifestyles; however, the impact on health behaviors including cancer prevention/screening behaviors has not been studied. Survivor-specific factors such as diagnosis, treatment, and chronic health conditions may also be associated with ongoing sibling psychologic distress (1, 22–26). As a result, they may serve as modifying factors for the development of adverse health behaviors including decreased cancer prevention/screening behaviors.

The purpose of this study was to (i) compare the cancer prevention and screening practices of siblings of childhood cancer survivors to the same practices in the general population and to (ii) identify demographic, health, and cancer-related factors associated with sibling cancer prevention and screening practices. To accomplish this, we used data from the Childhood Cancer Survivor Study (CCSS; refs. 27, 28) along with control data from the Behavioral Risk Factor Surveillance System (BRFSS) survey (29). We hypothesized that siblings of childhood cancer survivors would report decreased cancer screening and prevention practices in comparison with the general population. Furthermore, we hypothesized that survivor diagnosis, treatment intensity, and the presence of survivor late effects would be associated with a decrease in cancer prevention and screening practices.

Methods

Childhood Cancer Survivor Study

The CCSS is a retrospective cohort of long-term survivors of childhood cancer diagnosed between January 1, 1970, and December 31, 1986 and aggregated across 26 collaborating institutions in the United States and Canada. A description of the study design, methods, and sample used by the CCSS has been published previously (27, 28). Eligibility criteria for the CCSS include: (i) a diagnosis of leukemia, central nervous system malignancy, Hodgkin lymphoma, non-Hodgkin lymphoma, kidney tumor, neuroblastoma, bone tumor, or soft tissue sarcoma before 21 years of age and (ii) survival to at least 5 years postdiagnosis. Figure 1 provides a flow diagram of participants. Data for the study were collected between 2002 and 2004 via a 24-page self-report second follow-up questionnaire. Completed by both survivors and their siblings,

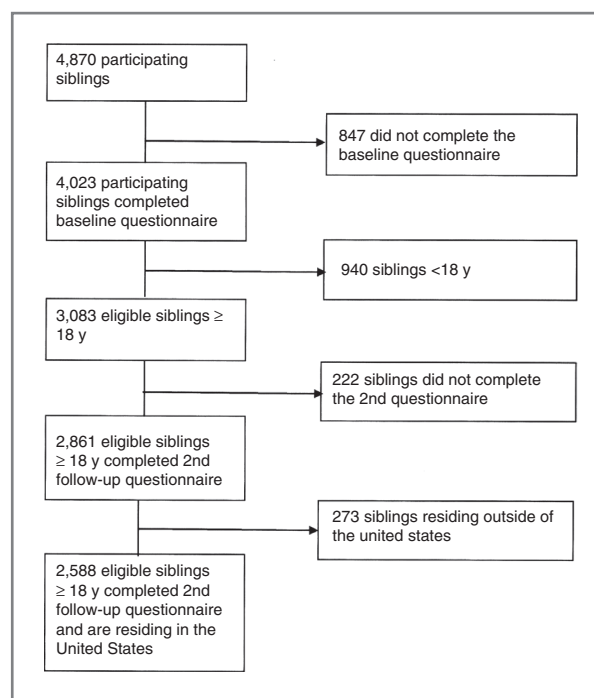


Figure 1. Flow diagram of participants.

the second follow-up questionnaire captured demographic information and information on physical and psychologic health. Following consent, treatment data were obtained from the survivors' treating institution by trained abstractors. The protocols and questionnaires underwent approval by the Institutional Review Boards of all collaborating centers and can be viewed at ccss.stjude.org.

Behavioral Risk Factor Surveillance System

As a comparison group, an age, gender, and race-matched sample of participants from the BRFSS survey (29) was selected in an approximate 2:1 (BRFSS:CCSS) ratio. The BRFSS is an annual, standardized, state-based, random-digit-dial telephone survey of the noninstitutionalized U.S. population completed with assistance from the Centers for Disease Control and Prevention. For the purpose of this study, data from the BRFSS survey administered in 2000 were used. The skin cancer module was administered in 6 states in 2000 including Colorado, Maryland, South Dakota, Virginia, Utah, and Washington. Breast and cervical cancer screening questions were part of the core questions administered in all states. Table 1 shows that cancer prevention and screening variables are closely comparable between the CCSS second follow-up questionnaire and the BRFSS survey administered in 2000. This similarity allows for assessment of screening and prevention practices across both populations at time points that ensure stability of cancer screening and prevention practices.

Table 1. Comparability of the CCSS and BRFSS cancer prevention and screening questions

CCSS	BRFSS
When you were outside last summer for more than 15 minutes how often did you protect yourself from the sun by applying sunscreen with a SPF 15 or more on all sun exposed skin areas?	When you go outside on a sunny summer day for more than 1 h, how often do you use sunscreen or sunblock?
When you were outside last summer for more than 15 minutes how often did you protect yourself from the sun by wearing protective clothing such as long-sleeved shirts and long pants?	When you go outside on a sunny summer day for more than an hour how often do you wear long-sleeved shirts?
When you were outside last summer for more than 15 minutes how often did you protect yourself from the sun by wearing a hat?	When you go outside on a sunny summer day for more than an hour how often do you wear a wide-brimmed hat or any other hat that shades your face, ears, and neck from the sun?
When you were outside last summer for more than 15 minutes how often did you protect yourself from the sun by staying in the shade?	When you go outside on a sunny summer day for more than an hour, how often do you stay in the shade?
When was the last time you had a mammogram?	How long has it been since your last mammogram?
When was the last time you had a Pap smear (test for cancer of the cervix)?	How long has it been since you had your last Pap smear?

Primary outcome measures

Skin cancer prevention behaviors. For both men and women, sun protection behaviors were the primary outcomes and included use of sunscreen, wearing protective clothing, wearing a hat, and staying in the shade. Five questions to assess this set of behaviors were similar across both the CCSS and the BRFSS as were response labels (CCSS: never, rarely, sometimes, often, and always; BRFSS: never, seldom, sometimes, near always, and always). Responses were recoded as a dichotomous outcome variable with those individuals that showed the practice (often/near always, always) and those that did not (never, rarely/seldom, sometimes) to provide a clinically meaningful expression of the results.

Breast and cervical cancer screening and prevention behaviors. For women, the other screening behavior outcomes included breast and cervical cancer screening, including mammography and Pap testing practices, and were questioned similarly between the CCSS and BRFSS. Responses for the CCSS (>1 year ago, 1–2 years ago, <2 years ago but >5 years ago, ≤5 years ago, do not know, and never) were similar to those for the BRFSS (within the past year or 1–12 months ago, within the past 2 years or 1–2 years ago, within the past 3 years or 2–3 years ago, within the past 5 years or 3–5 years ago, ≤5 years ago, do not know/not sure, refused). Responses were similarly recoded as a dichotomous outcome variable with those individuals that did not show the practice (≤5 years ago, do not know, never, or refused) and those that did (all other responses).

Independent variables

Sibling factors. Sociodemographic, health, and cancer-related variables were assessed as potential risk factors

for decreased cancer prevention/screening practices. Sibling sociodemographic factors included age at completion of the second follow-up questionnaire, sex, race/ethnicity, marital status, educational attainment, household income, employment status, and insurance status. Sibling health-related factors included self-reported psychologic health measured using the Brief Symptom Inventory 18 (BSI-18; refs. 30–32) and general health using a 5-category response scale: poor, fair, good, very good, and excellent. Sibling chronic health conditions were characterized according to the chronic health severity index derived from the NCI Common Terminology Criteria for Adverse Events (33). Chronic conditions of any grade were considered as a risk factor. Sibling cancer-related factors included sibling age at diagnosis of the survivor's cancer and presence or absence of sibling bereavement (i.e., whether or not the survivor died following study entry).

Survivor factors. Survivor factors included sociodemographic, health, and cancer-related variables. Sociodemographic factors included survivor age at the time of questionnaire completion and sex. The relative age of the survivor and the sibling was also considered (i.e., survivor older/sibling older). Survivor health-related factors included self-reported general and psychologic health measured as described above for siblings. Survivor cancer-related factors included diagnosis and treatment intensity as conceptualized previously using a yes/no composite variable of chemotherapy, surgery, and radiation therapy exposures (34). Late adverse outcomes of cancer and its treatment included chronic health conditions, defined as above for siblings, as well as development of a second cancer. For the outcomes of skin cancer prevention behaviors, the presence of a nonmelanoma skin cancer was also included.

Analysis

Data from the 2,588 siblings of the CCSS sample residing in the United States and 5,915 participants (for skin cancer prevention behavior analyses) as well as 37,789 female participants (for breast and cervical cancer screening practice analyses) from the BRFSS randomly drawn from an age, gender, and race-matched sample were analyzed. For the breast and cervical cancer screening sample, BRFSS females (all ages) were sampled from states that were excluded (i.e., states other than the 6 states that participated in the BRFSS skin cancer module) matching on age and race. This sample was added to the BRFSS sample used for the skin cancer prevention analyses. This combined BRFSS sample was used for the cervical cancer screening analyses; however, a subset of BRFSS females (i.e., greater than or equal to 40 years of age) was chosen for the breast cancer screening analyses.

Descriptive statistics were generated for sociodemographic, health, and cancer-related variables. The proportion of siblings and the proportion of BRFSS participants reporting individual cancer prevention/screening practice were calculated. Concordance between sibling and survivor cancer prevention/screening practices was also reported. Logistic regression was used to evaluate adjusted comparisons of cancer prevention and screening behaviors between the sibling sample and the selected participants from the BRFSS while controlling for age, sex, race/ethnicity, education, and income.

Within the sibling sample, multivariable logistic regression models were estimated for each individual cancer prevention/screening behavior. Candidate risk factors for the full model were chosen based on both results from an adjusted univariate analyses and the questions of interest. All models were adjusted for sibling age at completion of the second follow-up questionnaire, sibling sex, sibling race, sibling household income, sibling education, sibling health insurance. For models focusing on skin cancer prevention, region was included as an adjustment variable after classifying states, which administered the skin cancer module into regions (i.e., warm and cold regions; ref. 35). Each full model was reduced using likelihood ratio tests, eliminating variables of interest at the $\alpha = 0.05$ level. ORs and accompanying 95% confidence intervals (CI) are reported. We used SAS version 9.2 for all analyses.

Results

Provided in Table 2 are the characteristics of the sibling and BRFSS samples. The mean age at the time of questionnaire completion for the sibling population was 33.5 years (range, 18.0–58.0 years). The mean age of the sibling at the time of cancer diagnosis was 9.0 years (range, 10.1–36.3 years). Fifty-four percent of the siblings ($N = 1,392$) were born before their survivor and 46% of the siblings ($N = 1,196$) were born after their survivor. There was a mean interval of 25.1 years (range, 17.1–35.0 years) between the time of cancer diagnosis and sibling completion of the questionnaire.

Sibling and survivor concordance of cancer prevention and screening behaviors

The practice of sunscreen use was shared by both siblings and survivors 23.1% of the time. For protective clothing use, this practice was shared by 3.2% of the siblings and survivors. For hat use, siblings and survivors reported a similar commitment to this skin cancer prevention behavior 13.5% of the time. Shade use was reported by both siblings and survivors 11.7% of the time. In contrast, concordance between the practice of breast and cervical cancer screening practices of female siblings and survivors was higher. With respect to Pap testing, 82.2% of siblings and survivors report this behavior. Of those female siblings and survivors ≥ 40 years of age, concordance for the practice of mammography was 77.2%.

Adjusted analysis of cancer prevention and screening behaviors

Table 3 reports the frequencies of the cancer prevention and screening behaviors for the sibling and the BRFSS populations adjusted for age, sex, race/ethnicity, education, income, and region. The siblings were more likely to report skin cancer prevention behaviors including protective clothing use (OR, 2.85; 95% CI, 2.39–3.39), shade use (OR, 2.11; 95% CI, 1.88–2.36), sunscreen use (OR, 1.27; 95% CI, 1.14–1.40), and hat use (OR, 1.77; 95% CI, 1.58–1.98) compared with the BRFSS individuals after controlling for age, gender, race/ethnicity, education, income, and region ($P < 0.001$, all outcomes). No differences were noted with respect to cervical or breast cancer screening practices such as Pap testing ($P = 0.55$) or mammography ($P = 0.12$) after adjusting for age, race/ethnicity, education, and income.

Multivariable analysis for sibling cancer prevention and screening behaviors

Table 4 reports the multivariable analyses of factors impacting sibling skin cancer prevention behaviors. Being of male sex was consistently associated with skin cancer prevention behaviors as shown by increased use of clothing (OR, 1.69; 95% CI, 1.29–2.21) and increased use of hats (OR, 4.44; 95% CI, 3.55–5.59), but less use of sunscreen (OR, 0.59; 95% CI, 0.49–0.70) and shade (OR, 0.72; 95% CI, 0.59–0.87). Sibling age was also associated diminished practice of a subset of skin cancer prevention behaviors. Younger sibling age (i.e., 18–29 years) was associated with decreased protective clothing use (OR, 0.39; 95% CI, 0.19–0.82) and shade use (OR, 0.46; 95% CI, 0.27–0.79) when compared with siblings ≥ 50 years of age. Other sibling demographic factors predicting a subset of the skin cancer protective behaviors including being of white non-Hispanic race/ethnicity, which was associated with an increase in sunscreen use (OR, 2.00; 95% CI, 1.40–2.90), but less use of shade (OR, 0.60; 95% CI, 0.43–0.84).

Table 5 reports the adjusted analyses of factors impacting sibling breast and cervical cancer screening behaviors. Sibling health insurance was the only sibling factor

Table 2. Characteristics of the CCSS sibling populations and the age, gender, and race-matched random samples of participants from the BRFSS

	CCSS siblings (N = 2,588) Frequency (%)	BRFSS (N = 5,915) Frequency (%)	CCSS siblings (N = 1,388) Frequency (%)	BRFSS (N = 37,789) Frequency (%)
Age at interview, y				
18–19	82 (3.2)	187 (3.2)	44 (3.2)	1,209 (3.2)
20–29	849 (32.8)	1,961 (33.2)	436 (31.4)	11,875 (31.4)
30–39	966 (37.3)	2,220 (37.5)	536 (38.6)	14,548 (38.5)
40–49	605 (23.4)	1,358 (23.0)	315 (22.7)	8,582 (22.7)
≥50	86 (3.3)	189 (3.2)	57 (4.1)	1,575 (4.2)
Sex				
Female	1,388 (53.6)	3,176 (53.7)	1,388 (100.0)	37,789 (100.0)
Male	1,200 (46.4)	2,739 (46.3)		
Marital status				
Not married	1,004 (39.0)	2,295 (38.9)	503 (36.4)	14,985 (39.7)
Married/living as married	1,569 (61.0)	3,605 (61.1)	877 (63.6)	22,723 (60.3)
Employed				
Yes	2,179 (84.2)	4,781 (81.0)	1,088 (78.4)	27,045 (71.6)
No	409 (15.8)	1,125 (19.0)	300 (21.6)	10,701 (28.4)
Health insurance				
Yes	2,313 (89.7)	5,897 (99.7)	1,254 (90.5)	37,724 (99.8)
No	267 (10.3)	18 (0.3)	131 (9.5)	65 (0.2)
Race/ethnicity				
Other	204 (8.2)	488 (8.3)	114 (8.5)	3,206 (8.5)
White non-Hispanic	2,296 (91.8)	5,427 (91.7)	1,229 (91.5)	34,583 (91.5)
Household income				
0–\$19,999	240 (9.3)	682 (12.7)	145 (10.4)	6,196 (18.5)
≥\$20,000	2,348 (90.7)	4,698 (87.3)	1,243 (89.6)	27,344 (81.5)
Education				
Less than high school	57 (2.2)	375 (6.4)	26 (1.9)	2,954 (7.8)
High school graduate	321 (12.4)	1,627 (27.6)	152 (11.0)	11,074 (29.3)
Greater than high school	2,203 (85.4)	3,901 (66.1)	1,206 (87.1)	23,722 (62.8)
Survivor diagnosis				
Leukemia	902 (34.8)		476 (34.3)	
Central nervous system	321 (12.4)		166 (12.0)	
Hodgkin disease	372 (14.4)		211 (15.2)	
Non-Hodgkin lymphoma	202 (7.8)		116 (8.4)	
Kidney	222 (8.6)		124 (8.9)	
Neuroblastoma	159 (6.1)		75 (5.4)	
Soft tissue sarcoma	220 (8.5)		115 (8.3)	
Bone cancer	190 (7.3)		105 (7.6)	
Survivor's treatment intense				
Yes	1,851 (77.2)		988 (76.7)	
No	546 (22.8)		301 (23.3)	

associated with the practice of both breast and cervical cancer screening. The presence of health insurance was associated with greater use of Pap testing (OR, 2.11; 95% CI, 1.13–3.80) and greater use of mammography (OR, 6.36; 95% CI, 1.40–28.6). Being a sibling of white non-Hispanic race/ethnicity was associated with greater Pap testing.

With few exceptions, there were no significant associations between the survivor's diagnosis, the intensity of the

survivor's treatment, the presence or absence of a chronic health condition in the survivor, psychologic distress in the survivor, second cancers, across skin cancer prevention behaviors as well as breast, and cervical cancer screening practices. A survivor diagnosis of a sarcoma or a bone tumor was associated with greater use of protective clothing and hat use among siblings when compared with a survivor diagnosis of leukemia. Fair/poor survivor

Table 3. Percentages of CCSS siblings and the age, gender, and race-matched sample from BRFSS reporting cancer prevention and screening behaviors as well as adjusted odds ratios for each category

Cancer prevention and screening behavior	CCSS siblings N (%)	BRFSS N (%)	Adjusted OR (CCSS vs. BRFSS) ^a OR (95% CI)
Protective clothing use			
Yes	343 (13.4)	318 (5.5)	2.85 (2.39–3.39) ^b
No	2,212 (86.6)	5,458 (94.5)	
Staying in shade			
Yes	805 (31.5)	1,076 (18.8)	2.11 (1.88–2.36) ^b
No	1,753 (68.5)	4,645 (81.2)	
Sunscreen use			
Yes	1,092 (42.5)	1,965 (34.0)	1.27 (1.14–1.40) ^b
No	1,475 (57.5)	3,818 (66.0)	
Wearing a hat			
Yes	873 (34.3)	1,371 (23.7)	1.77 (1.58–1.98) ^b
No	1,675 (65.7)	4,407 (76.3)	
Pap smear			
Yes	1,273 (93.1)	34,321 (91.5)	0.94 (0.76–1.17)
No	95 (6.9)	3,187 (8.5)	
Mammogram ^c			
Yes	312 (85.2)	8,018 (79.5)	1.27 (0.95–1.73)
No	54 (14.8)	2,069 (20.5)	

^aAdjusted for age, gender, race/ethnicity, education, income, and region.

^b $P \leq 0.001$.

^cOnly females >40 years of age were included.

health status was associated with greater use of protective clothing and Pap testing among siblings. The presence of a nonmelanoma skin cancer in the survivor was not associated with skin cancer prevention behaviors in the siblings.

Discussion

To our knowledge, this is the first study of siblings of adult childhood cancer survivors to examine the impact of the childhood cancer experience on sibling cancer prevention and screening practices. Contrary to our hypothesis, we show that, as a group, adult siblings of long-term survivors of childhood cancer report greater practice of skin cancer prevention practices when compared with the general population. In female siblings there was no significant difference in breast and cervical cancer screening. We also identified various factors associated with health risk and health protective behaviors. Being a sibling with less than a high school education and lacking health insurance was associated with diminished skin cancer prevention behaviors (i.e., sunscreen use) as well as diminished breast and cervical cancer screening practices, respectively.

Skin cancer prevention behaviors

Greater skin cancer prevention behaviors in siblings of childhood cancer survivors were showed when compared

with the general population. Despite the stress associated with the childhood cancer experience and hypothesized development of adverse adult health behaviors as a result, the ability of siblings to positively adapt and cope with the cancer experience may be significant. Research has documented that sibling's experience fear of developing cancer themselves (5–8) which may be associated with greater perceived risk of cancer or commitment to skin cancer prevention on the part of the sibling. Sibling's higher rates of posttraumatic growth (36) may also serve to better equip them to face their own risk for cancer by using less avoidant coping strategies or denial to circumvent these emotional responses. As a result, siblings may have a greater perceived vulnerability to cancer and commitment to practice skin cancer prevention behaviors. Siblings of melanoma survivors show a similar pattern of greater skin cancer prevention behaviors than the general population (37). Ongoing parental and family distress may also result in the recommendations of family members for siblings to focus on cancer prevention and screening (38).

Sibling demographic factors including sibling sex and race/ethnicity were associated with increases in specific cancer prevention behaviors and decreases in others. Male siblings showed decreased sunscreen and shade use in addition to increased use of protective clothing and hats. Research in community samples has showed decreased

Table 4. Multivariable models for sibling skin cancer prevention behaviors

Sibling and survivor factors	Sunscreen use OR (95% CI)	Protective clothing OR (95% CI)	Wearing hat OR (95% CI)	Shade OR (95% CI)
Sibling sex				
Male	0.59 (0.49–0.70) ^b	1.69 (1.29–2.21) ^b	4.44 (3.55–5.59) ^b	0.72 (0.59–0.87) ^b
Female (referent)	1.00	1.00	1.00	1.00
Sibling age at study, y				
18–29	0.63 (0.37–1.07)	0.39 (0.19–0.82) ^a	0.59 (0.31–1.17)	0.45 (0.27–0.78) ^a
30–39	0.90 (0.53–1.52)	0.52 (0.27–1.07)	0.80 (0.42–1.54)	0.70 (0.41–1.21)
40–49	1.04 (0.61–1.79)	0.53 (0.27–1.09)	1.03 (0.55–1.99)	0.73 (0.42–1.26)
≥50 (referent)	1.00	1.00	1.00	1.00
Race/ethnicity				
White (non-Hispanic)	2.00 (1.40–2.90) ^b	1.14 (0.70–1.93)	1.22 (0.78–1.94)	0.60 (0.43–0.83) ^a
Other (referent)	1.00	1.00	1.00	1.00
Household income				
0–\$19,999	0.89 (0.64–1.24)	0.84 (0.50–1.37)	1.15 (0.76–1.72)	1.40 (1.01–1.93) ^a
\$20,000+ (referent)	1.00	1.00	1.00	1.00
Education				
Less than high school	0.46 (0.21–0.92) ^a	1.38 (0.54–3.12)	0.91 (0.41–1.95)	0.48 (0.20–1.02)
High school graduate	0.48 (0.36–0.64) ^b	1.26 (0.85–1.84)	0.77 (0.54–1.08)	1.04 (0.79–1.37)
Greater than high school (referent)	1.00	1.00	1.00	1.00
Health insurance				
Yes	1.59 (1.16–2.21) ^a	0.76 (0.50–1.18)	0.96 (0.66–1.42)	0.91 (0.67–1.25)
No (referent)	1.00	1.00	1.00	1.00
Region (warm state)				
Yes	1.26 (1.05–1.51) ^a	1.22 (0.93–1.61)	1.47 (1.17–1.85) ^b	1.05 (0.86–1.27)
No (referent)	1.00	1.00	1.00	1.00
Sibling psychologic distress ^c				
Yes				1.49 (1.01–2.17) ^a
No (referent)				1.00
Survivor diagnosis				
Brain tumor		0.91 (0.55–1.45)	0.79 (0.53–1.17)	
Hodgkin lymphoma		1.11 (0.70–1.72)	1.24 (0.86–1.78)	
Non-Hodgkin lymphoma		1.59 (0.97–2.54)	1.54 (1.00–2.36) ^a	
Kidney tumor		0.63 (0.34–1.12)	1.51 (0.99–2.29)	
Neuroblastoma		1.60 (0.92–2.69)	1.50 (0.91–2.44)	
Soft tissue sarcoma		1.80 (1.12–2.86) ^a	1.33 (0.86–2.04)	
Bone tumor		0.83 (0.45–1.47)	1.63 (1.04–2.54) ^a	
Leukemia (referent)		1.00	1.00	
Survivor psychologic distress ^c				
Yes			0.57 (0.38–0.86) ^a	
No (referent)			1.00	
Survivor chronic health conditions				
Yes				1.25 (1.01–1.55) ^a
No (referent)				1.00
Survivor health				
Fair/poor		1.53 (1.04–2.19) ^a		
Excellent/very good/good (referent)		1.00		

NOTE: Sibling and survivor factors not significant ($P \geq 0.1$) were not included in final model aside from sibling sex, sibling age at study, race/ethnicity, income, education, and region.

^a $P \leq 0.05$.

^b $P \leq 0.001$.

^cPsychologic distress = GSI for the outcome of psychologic distress.

Table 5. Multivariable models for sibling cervical and breast cancer prevention and screening behaviors

Sibling factors	Pap test OR (95% CI)	Mammogram ^c OR (95% CI)
Sibling age at study, y		
18–29 ^d	0.67 (0.10–2.44)	
30–39 ^d	1.59 (0.24–5.99)	
40–49	0.88 (0.13–3.40)	0.70 (0.20–1.98)
≥50 (referent)	1.00	1.00
Race/ethnicity		
White (non-Hispanic)	3.61 (1.97–6.40) ^b	1.01 (0.20–3.73)
Other (referent)	1.00	1.00
Household income		
0–\$19,999	1.06 (0.54–2.27)	5.72 (0.92–119.94)
\$20,000+ (referent)	1.00	1.00
Education		
Less than high school	0.28 (0.09–1.08) ^a	
High school graduate	0.40 (0.23–0.72) ^a	
Greater than high school (referent)	1.00	
Health insurance		
Yes	2.11 (1.13–3.80) ^a	6.36 (1.40–28.6) ^a
No (referent)	1.00	1.00
Sibling chronic health conditions		
Yes		0.27 (0.07–0.75) ^a
No (referent)		1.00
Survivor health		
Fair/poor	1.68 (1.05–2.73) ^a	0.44 (0.20–1.04)
Excellent/very good/good (referent)	1.00	1.00
Birth order		
Survivor younger	2.01 (1.21–3.42) ^a	
Survivor older (referent)	1.00	

NOTE: Sibling and survivor factors not significant ($P \geq 0.1$) were not included in final model aside from sibling sex, sibling age at study, race/ethnicity, income, and education.

^a $P \leq 0.05$.

^b $P \leq 0.001$.

^cOnly females >40 years of age were included.

^dThese age categories were used only for the outcome of Pap testing.

sunscreen use in males (39) and increased use of protective clothing (40). Gender differences may relate to the fact that women tend to sunbathe more when sunscreen may also be used and men tend to be more likely to have occupational exposure to sun when protective clothing is more likely to be used. White non-Hispanic siblings endorsed greater sunscreen use, but less shade use. Research with nononcology groups has also showed that white non-Hispanic individuals show greater sunscreen use than other racial and ethnic groups (39).

Other sociodemographic factors including sibling age, educational attainment, and health insurance status were associated with skin cancer prevention practices. Sibling age at the time of study was associated with skin cancer prevention practices among siblings. Younger sibling age was also associated with diminished skin cancer prevention behaviors. In the general

population, older individuals show greater knowledge of skin cancer prevention behaviors and more skin cancer prevention practices (41). Similar to the general population, siblings with less educational attainment may have decreased knowledge of, and subsequent practice of, skin cancer prevention behaviors including sunscreen use. Research has showed that knowledge of skin cancer prevention practices is an important predictor of these behaviors (42). Furthermore, modifiable factors such as knowledge have been the basis of many interventions aimed at improving skin cancer prevention practices in other at-risk populations (37). Siblings with health insurance endorsed greater prevention behaviors including sunscreen use. Health insurance is associated with improved access to care and greater likelihood of participating in cancer prevention behaviors (43).

Breast and cervical cancer prevention and screening behaviors

The practice of breast and cervical cancer prevention behaviors in siblings is similar when compared with the general population. Despite this similarity, these figures fall short of the cancer screening goals set forth in Healthy People 2020 (44). As mentioned previously, siblings of colorectal cancer survivors (45, 46) and melanoma survivors (37) report greater cancer prevention practices than siblings of noncancer survivors. In contrast, our comparison of siblings of survivors to the general population suggests that the childhood cancer experience appears to have little impact on the breast and cervical cancer screening practices of siblings. It is possible that attitudes toward risk and perceptions of risk among female siblings may mediate the impact of the cancer experience on sibling health behaviors including breast and cervical cancer screening. The greater concordance between siblings and survivors with respect to breast and cervical cancer screening practices when compared with skin cancer prevention practices provides additional support for this hypothesis; however, additional data will be necessary to test this hypothesis.

Overall sibling health insurance status was associated with breast and cervical cancer screening practices among siblings. The presence of sibling health insurance was also associated with greater Pap testing and mammography use. As significant shifts have occurred in the delivery of health care within the United States, adherence to recommended screening guidelines such as those published by various health organizations including the American Cancer Society (e.g., yearly mammography beginning at 40 years of age and Pap testing beginning no later than 21 years of age; ref. 47) are important markers of the delivery of necessary health services. Sibling health behaviors including the practice of cancer screening practices such as Pap testing and mammography are likely to be influenced by the childhood cancer experience; however, health insurance status remains a significant predictor of the delivery of these services to a population at-risk for the development of cancer (43).

Sociodemographic factors including sibling educational attainment and race/ethnicity were associated with breast and cervical cancer screening practices. Similar to the general population, siblings with less educational attainment may have decreased knowledge of and subsequent practice of Pap testing. Knowledge of cancer prevention and screening is an important predictor of cancer screening behaviors including Pap testing (48). Furthermore, educational interventions aimed at improving Pap testing use in other at-risk populations has been described (49). White non-Hispanic siblings endorsed greater Pap testing. Research with nononcology groups has showed that white non-Hispanic individuals have showed superior cervical cancer screening rates; however, the gaps between groups have decreased (50).

Survivor diagnosis, treatment intensity, and the presence of survivor late effects including adverse general and

psychologic health, chronic health conditions, and second cancers, did not emerge as salient predictors of sibling cancer prevention/screening practices. Being a sibling of a survivor with a sarcoma or a bone tumor compared with leukemia was associated with greater use of protective clothing and hats, respectively. Disfigurement and persistent hair loss is common for childhood cancer survivors and associated with ongoing survivor distress (51). Adverse survivor health was also associated with greater use of protective clothing and Pap testing. These ongoing familial reminders of the cancer experience may be associated with perception of cancer risk and greater practice of skin cancer prevention behaviors among siblings. These issues are clinically important; however, these analyses did not uncover overwhelming evidence of relationships between these variables and cancer prevention behaviors among siblings. Of particular note, a diagnosis of the most common second cancer in the survivor, a nonmelanoma skin cancer, (52) did not impact skin cancer prevention behaviors among siblings.

When interpreting the results of our study, the following limitations should be considered. Differences exist between the BRFSS and the CCSS including the timing of the individual studies (2000 vs. 2002–2004) and the time of exposure of the skin cancer prevention questions (1 hour vs. 15 minutes). We anticipate that the difference in time of exposure would elicit a bias toward decreased skin cancer prevention behaviors in the siblings. Despite sample differences, attempts were made to address differences by controlling for factors such as age, sex, education, and income. The sociodemographic characteristics of the CCSS cohort also need to be considered. There may be biases introduced by the fact that not all of the siblings of the randomly selected survivors participated and, compared with survivors, the sibling sample overrepresents white non-Hispanic families. However, in regards to race/ethnicity, the sibling sample was closely matched to the BRFSS resulting in reliable estimates in health behaviors where race/ethnicity could be a factor. Despite these limitations, this is the largest collection of siblings of long-term survivors of childhood cancer. The CCSS cohort provides a similar socioeconomic cohort of survivors and nearest age siblings so that examination of within family, sibling, and survivor factors can be analyzed for the impact of the cancer experience on siblings' health behaviors.

In summary, siblings endorse the practice of skin cancer prevention and screening practices to a greater extent than the general population and breast and cervical cancer screening practices at a level equal to the general population. These results support a modified hypothesis in which despite the stress induced by the childhood cancer experience, siblings are able to show positive adaptation as manifest by the practice of health protective behaviors. We found that very few of the hypothesized predictors were consistently associated across cancer prevention and screening behaviors. Therefore, these predictors are inadequate in their

ability to define at-risk siblings in need of intervention for problematic health behaviors. The findings support the need to develop a better understanding of those factors, which support health promotion and minimize health risk behaviors (e.g., access to care and education) among siblings of childhood cancer survivors. Such an investment would inform the development of effective interventions to optimize health behaviors among siblings of childhood cancer survivors.

Disclosure of Potential Conflicts of Interest

No potential conflicts of interest were disclosed.

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