

Financial hardship among siblings of long-term survivors of childhood cancer: a Childhood Cancer Survivor Study (CCSS) report.

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BACKGROUND & RATIONALE:

Approximately 500,000 survivors of childhood cancer reside in the United States, many of whom are at elevated risk for physical, neurocognitive, and psychosocial morbidities related to their cancer and treatment.¹⁻³ It is well known that adult survivors of pediatric cancer experience poorer health outcomes compared to the general population,⁴ but greater attention in recent years has been given to the financial wellbeing of survivors of pediatric cancer, identifying areas of concern.⁵⁻⁷ Survivors of childhood cancer incur significantly greater medical expenditures, obtain lower-paying employment, and miss more days of work resulting in productivity loss.⁸⁻¹⁰ Accumulation of these difficulties may place survivors at increased risk for various types of financial hardship: material [difficulty in affording basic necessities or medical care], psychological [excessive worry about finances], and/or behavioral [intentionally avoiding or rationing recommended medical care to save money].^{5,11,12} In survivors of adult-onset cancers, greater financial burden has been shown to be associated with poorer health-related quality of life and inferior survival.^{13,14}

Recent investigation from the CCSS demonstrated that survivors report a greater incidence of various financial hardships as compared to sibling controls.^{15,16} In this effort to more fully understand the burden of financial hardship, the CCSS surveyed a large, geographically diverse cohort of childhood cancer survivors, along with sibling controls, with respect to a range of financial measures. This survey, titled Follow-up 6 medium (FU-6), was developed with several questions derived from the National Health Interview Survey (NHIS; 11 questions),¹⁷ Behavioral Risk Factor Surveillance System (BRFSS; specifically 3 questions adapted from the Social Determinants of Health section),¹⁸ and Medical Expenditure Panel Survey (MEPS; 8 questions from the general survey and 2 questions from the Special Experiences with Cancer section).¹⁹ The BRFSS Social Determinants of Health section was a supplemental survey administered in

17 states in 2017. A landmark comparison of cancer survivors to age-matched siblings by Nathan and colleagues (manuscript currently in review) demonstrated that survivors experienced a significantly greater burden of financial hardship across multiple domains.¹⁶

However, the financial experiences of siblings of children with cancer are unknown. Prior research has demonstrated that cancer treatment has marked impact on siblings in several domains, given their proximity, emotional ties, and shared household resources during childhood.²⁰⁻²² For example, emotional impairment is common among siblings during treatment and often persists following therapy completion.²³⁻²⁷ It is also possible that adult siblings may devote significant time and financial resources to caregiving or support for survivors with ongoing medical needs, although this is not well studied.²⁸ The existing literature in similar psychosocial domains, as well as the potential impact of cancer treatment, provides strong rationale to examine financial hardship specifically in siblings.¹⁶ One of the strengths of the CCSS is the availability of age-matched siblings as controls;^{29,30} however, while Nathan and colleagues identified key financial challenges experienced by cancer survivors compared with siblings, the use of sibling controls was also cited as a potential limitation, given the known overlap in their psychosocial outcomes.¹⁶ A comparison of siblings to the general population of adults in the U.S. may improve the understanding of the financial hardship experienced by siblings, and also strengthen the prior survivor-sibling comparisons made by Nathan et al., particularly if differences between siblings and the general population are noted. Furthermore, identification of risk factors for adverse financial outcomes in siblings may identify areas for further investigation or intervention for both cancer survivors and their family members.

To evaluate the effect of cancer treatment on financial hardship experienced by siblings, we seek to describe the prevalence of domains of financial hardship in this group and compare the prevalence of various aspects of financial hardship in siblings directly to the general population of adults without a cancer history. We will also examine characteristics within the sibling group itself that may be associated with greater prevalence of types of financial hardship. Our outlined aims will use individual items of financial hardship on FU-6 to the original questions from the NHIS and BRFSS for comparisons between siblings and the general population, and will calculate domain scores of types of financial hardship (material, psychological, and behavioral), previously established by Nathan, et al,¹⁶ for comparisons within sibling groups.

OVERVIEW OF AIMS/SURVEYS:

Aim 1: Describe the prevalence of financial hardship in siblings of childhood cancer survivors. The reported variables of interest will be items obtained from FU-6. Outcome variables are listed in Table 1, with a summary of criteria to determine a positive response to each item. This aim is descriptive, so there are no hypotheses.

Aim 2: Compare the prevalence of financial hardship in siblings of childhood cancer survivors with adults from the general population without a cancer history. Variables of interest are matching items obtained from FU-6 and corresponding national surveys, as summarized in Table 2.

- Hypothesis: *Siblings of childhood cancer survivors will report a higher prevalence of indicators financial hardship when compared with the general population.*

Aim 3: Identify characteristics associated with greater financial hardship in siblings of cancer survivors. Outcome variables of interest will be obtained from FU-6 and are summarized in Table 1, with domain scores calculated per Table 3. Sibling characteristics of interest include sibling sociodemographic and health status-related factors, as well as their family member's (i.e., cancer survivor) disease and treatment-related factors. Exposure variables and covariates are summarized in Table 4.

- Hypothesis: *Siblings with certain sociodemographic characteristics, as well as chronic medical conditions, will experience greater financial hardship compared with siblings without these factors. Furthermore, siblings of cancer survivors who received more intensive therapy, such as those with hematopoietic cell transplant or late relapse, will report a greater prevalence of financial hardship compared with siblings without these treatment-associated factors.*

ANALYSIS FRAMEWORK:

Populations of interest:

The current proposal will use financial hardship data from the CCSS FU-6 survey of cancer survivors and siblings from the CCSS, which was collected between 2017-2019, as well as data from the 2019 National Health Interview Survey (NHIS), publicly available at (<https://www.cdc.gov/nchs/nhis/2019nhis.htm>). The 2019 NHIS was specifically selected for comparison because it contained identical questions to some of FU-6 and was also conducted synchronously. Furthermore, the 2019 NHIS also received a substantial overhaul from prior iterations, including an updated sample weighting approach.³¹ Data from the 2017 Behavioral Risk Factor Surveillance System (BRFSS) will also be examined in an exploratory analysis (https://www.cdc.gov/brfss/annual_data/annual_2017.html). The BRFSS analysis will be exploratory because the BRFSS items did not correspond exactly with FU-6, and there were a small number of overlapping elements overall. As CCSS participants were all under age 65, national surveys will include responses only from adults under age 65. Items matching the MEPS will not be examined as these questions evaluated baseline factors such as income and assets, rather than outcomes related to financial hardship. Additionally, some items from the NHIS pertaining to income, debt, and assets will not be examined in the comparison aim.

Outcomes of interest:

Individual items for all aims are shown in Table 1, with criteria for response dichotomization. For Aim 2, individual items pertaining to financial hardship were selected from FU-6 for comparison to the general population if there was a parallel question available from the NHIS (7 questions, shown in Table 2) or BRFSS (2 questions, Table 2). Items from FU-6 used for analysis matched those from the NHIS verbatim. Items from FU-6 matched the BRFSS, with some small exceptions that are detailed in Table 2. Both the NHIS and BRFSS contain information regarding race, ethnicity, age, educational attainment, and marital status, factors that will be available for weighting in order to facilitate comparison to the CCSS FU-6 survey respondents. More description and rationale of the sample weighting process is below.

For Aim 3, domain scores will be calculated from selected outcomes of interest in Table 1, as shown in Table 3. The construction of these domain scores (material, psychological, and behavioral) will be identical to the model used by Nathan et al. Some additional questions will also be considered for analysis outside of the three financial hardship domains.

Table 1: Individual financial hardship items of interest from FU-6.

CCSS variable	Question	Response positive for financial hardship	Response negative for financial hardship
C4	How concerned are you that you may not be able to maintain your current level of insurance coverage over the next 12 months?	Moderately concerned, greatly concerned	Not concerned, a little concerned, missing
C5	In the past 12 months, did you/anyone in the family have problems paying or were unable to pay any medical bills? Include bills for doctors, dentists, hospitals, therapists, medication, equipment, nursing home or home?	Yes	No, don't know, missing
C6	Do you or anyone in your family currently have medical bills that you are unable to pay at all?	Yes	No, don't know, missing
C7	Do you are anyone in the house you live in have medical bills that are being paid off over time?	Yes	No, missing
C8	During the past 12 months, was there a time when you needed one of the following, but did not get it because you couldn't afford it? Check all that apply.		
C8a	Any needed medical care	Yes	No, don't know, missing
C8b	Yearly visit to your PCP	Yes	No, don't know, missing
C8c	Prescription medicine	Yes	No, don't know, missing
C8d	Mental health care or counseling	Yes	No, don't know, missing
C8e	Dental care	Yes	No, don't know, missing
C8f	Eye care	Yes	No, don't know, missing
C8g	Care from a specialist	Yes	No, don't know, missing
C9	Have you or your family had to make any other kinds of financial sacrifices in the past 2 years because of debt related to medical care? Check all that apply.	Yes to any subsection	
C9a	Reduced spending on vacation or leisure activities	Yes	No, missing
C9b	Reduced spending on purchasing large items (e.g. a car)	Yes	No, missing
C9c	Reduced spending on basics (e.g. food and clothing)	Yes	No, missing
C9d	Delayed or reduced spending on home improvement	Yes	No, missing
C9e	Used savings set aside for other purposes (e.g. retirement, educational funds, family support)	Yes	No, missing
C9f	Made a change to living situation (e.g. sold, refinanced or moved to a smaller residence).	Yes	No, missing
C10	During the past year, about how much did you spend out-of-pocket for your medical care?	≥10% of annual income	<10% of annual income
C11	Has your physical condition or medical treatment caused you financial difficulties over the past week?	A little difficulty, Quite a bit of difficulty, Great difficulty	No difficulty, missing
C12	Has your physical condition or medical treatment caused you financial difficulties over the past year?	A little difficulty, Quite a bit of difficulty, Great difficulty	No difficulty, missing
C13	In the past 2 years, were you working for pay either full time or part time at a job or business?	Yes	No, don't know, missing
C14	In the past 2 years, did you ever:		
C14a	Take extended paid time off from work, unpaid time off, or make a change in your hours, duties or employment status?	Yes	No, missing

C14b	Change to a less demanding job?	Yes	No, missing
C14c	Change from a set work schedule, where you start and end at the same the time every day, to a flexible work schedule, where your start and end times vary from day to day?	Yes	No, missing
C14d	Decide not to pursue an advancement or promotion?	Yes	No, missing
C14e	Worry that you might be forced to retire or quit work before you are ready?	Yes	No, missing
C14f	Stay at a job in part because you were concerned about losing your health insurance?	Yes	No, missing
C15	Did your spouse or significant other ever stay at a job in part because he/she was concerned about losing health insurance for the family?	Yes	No, missing
C16	If you get sick or have an accident, how worried are you that you will not be able to pay your medical bills?	Very worried, somewhat worried	Not worried, missing
C17	How often in the last 12 months would you say you were worried or stressed about having enough money to pay your rent or mortgage?	Always, usually, sometimes, rarely	Never, don't know, missing
C18	How often in the last 12 months would you say you were worried or stressed about having enough money to buy nutritious meals?	Always, usually, sometimes, rarely	Never, don't know, missing
C19	How often in the last 12 months would you say you were worried or stressed about having enough money to pay household utilities, including water, gas, and electricity?	Always, usually, sometimes, rarely	Never, don't know, missing
C29	Have you ever been sent to collections because of debts you were unable to pay on time or at all?	Yes	No, missing
C30	Have you ever filed for bankruptcy because of debts you were unable to pay?	Yes	No, missing

Table 2: Individual items pertaining to financial hardship to be used for sibling-general population comparisons.

FU-6 Variable	Question	NHIS Variable	BRFSS Variable
C5	In the past 12 months, did you/anyone in the family have problems paying or were unable to pay any medical bills? Include bills for doctors, dentists, hospitals, therapists, medication, equipment, nursing home or home?	PAYBLL12M_A	
C6	Do you or anyone in your family currently have medical bills that you are unable to pay at all?	PAYNOBLLNW_A	
C8	During the past 12 months, was there a time when you needed one of the following, but did not get it because you couldn't afford it? Check all that apply.		
C8a	Any needed medical care	MEDNG12M_A	
C8c	Prescription medicine	RXSK12M_A	
C8d	Mental health care or counseling	MHTHND_A	
C8e	Dental care	DENNG12M_A	
C16	If you get sick or have an accident, how worried are you that you will not be able to pay your medical bills?	PAYWORRY_A	
C18	How often in the last 12 months would you say you were worried or stressed about having enough money to buy nutritious meals?		Module 24 Q5*(SDHMEALS)

C17	How often in the last 12 months would you say you were worried or stressed about having enough money to pay your rent or mortgage?		Module 25 Q1* (SDHBILLS)
C19	How often in the last 12 months would you say you were worried or stressed about having enough money to pay household utilities, including water, gas, and electricity?		

*All questions from FU-6 were identical to the NHIS questions. The BRFSS question corresponding to these items stated: "During the last 12 months, was there a time when you were not able to pay your mortgage, rent or utility bills?" For comparison purposes to FU-6, questions C17 and C19 will be combined to compare to a single BRFSS item. A positive response to either item in FU-6 will be counted as positive for financial hardship. An answer of "Yes" to either of the two BRFSS items will be counted as positive for financial hardship.

Table 3: Domain score calculation: sum of responses to individual items, not factor scores.

Hardship domain	Items comprising domain
Behavioral	C8a, C8b, C8c, C8d, C8e, C8f, C8g
Material/financial sacrifices	C5, C7, C9a, C9b, C9c, C9d, C9e, C9f
Psychological hardship	C17, C18, C19

Exposures and covariates of interest:

Additional variables to be obtained from the CCSS and national surveys are shown in Table 4, with details of variable coding for each question from national survey data.

Table 4: Demographics, chronic health conditions, treatment exposures, and other covariates.

Measure	Data Source	CCSS Definition	NHIS/BRFSS Variables
Sibling-specific covariates			
Age	CCSS/NHIS/BRFSS data	Age at time of questionnaire	BRFSS: _AGEG5YR NHIS: AGE1_A or AGE2_A
Sex	CCSS/NHIS/BRFSS data	Male; female; other;	BRFSS: SEX NHIS: SEX_A
Race	CCSS/NHIS/BRFSS data	White; Black; American Indian or Alaska Native; Asian or Pacific Islander; Other, specify	BRFSS: _MRACE1 NHIS: RACEALLP_A
Ethnicity	CCSS/NHIS/BRFSS data	Hispanic; Non-Hispanic	BRFSS: _HISPANC NHIS: HISP_A
Education	CCSS/NHIS/BRFSS data	<High school; High school-some college; College graduate or greater	BRFSS: EDUCA NHIS: EDUC_A
Marital status	CCSS/NHIS/BRFSS data	Married; Single; Divorced or separated	BRFSS: MARITAL NHIS: MARSTAT_A
Household income	FU-6 C21, NHIS/BRFSS data	Household income: Less than \$20,000; \$20,000-39,999; \$40,000-59,999; \$60,000-79,999; \$80,000-99,999; \$100,000 or more; Don't know; Prefer not to answer	BRFSS: INCOME2 NHIS: INCGRP_A NHIS restricted portion: INCTOTAL_A

Chronic conditions	CCSS/NHIS/BRFSS data	None; 1 chronic condition; Multiple chronic conditions	Both surveys can ascertain this with similar lists of variables
Insurance coverage	FU-6 C1 and C2	Yes (Individual insurance, employer-based, Medicare, Medicaid, IHS, military, Other state-sponsored, Other government program, Don't know); No	
Insurance deductible	FU-6 C3	Greater than \$2,600; \$1,301-2600; Less than \$1,300; Don't know; missing	
Out-of-pocket expenses in last year	FU-6 C10	≥10% of annual income; <10% of annual income	
Homeownership	FU-6 C22, C23	Yes; No	
Value of home	FU-6 C24	\$0-\$100,000; \$100,001-\$250,000; \$250,001-\$500,000; \$500,001 or more; Don't know; Prefer not to answer	
Outstanding mortgage on home	FU-6 C25	Yes; No; Don't know	
How much owed on home	FU-6 C26	\$0-\$50,000; \$50,001-\$100,000; \$100,001-\$250,000; \$250,001-\$500,000; \$500,001 or more; Don't know; Prefer not to answer	
Other debts (credit cards, car loans, etc.)	FU-6 C27	Yes; No; Don't know	
Amount owed on other debt	FU-6 C28	\$0-\$25,000; \$25,001-\$50,000; \$50,001-\$100,000; \$100,001-\$250,000; \$250,001-\$500,000; \$500,001 or more; Don't know; Prefer not to answer	
Cancer survivor-specific covariates*			
Age at diagnosis	CCSS data	0-4.99, 5-9.99, 10-14.99, 15-20 years	
Time elapsed from diagnosis	CCSS data	10-19.99, 20-29.99, 30-39.99, ≥40 years	
Sex	CCSS data	Female; Male	
Cancer diagnosis	CCSS data	Leukemia; CNS tumor; HL; NHL; Wilms tumor; Neuroblastoma; Soft tissue sarcoma; Bone cancer	
Chronic conditions	CCSS/NHIS/BRFSS data	None; 1 chronic condition; Multiple chronic conditions	
Radiation	CCSS data	None; TBI only; Cranial RT, no TBI; Chest RT and without Cranial TBI; Other RT	
Surgery only (without chemo/radiotherapy)	CCSS data	Yes; No	
Stem cell transplant	CCSS data	Yes; No	
History of late relapse	CCSS data	Yes; No	

*Data would be limited to cancer survivors from the same family as siblings who participated in FU-6

Methods and data analysis:

General considerations: Because the selected items for comparisons to national surveys do not comprehensively cover the FU-6 survey, domain scores based on material, psychological, or behavioral hardship will not be calculated Aim 2; rather, responses to items will be compared individually between the CCSS and national survey data. A matching question from the BRFSS covering household material hardship related to rent and utilities will be separated into two questions on the FU-6 as shown in Table 2. Composite responses to both FU-6 questions will

be compared to this individual BRFSS item. For Aim 3, domain scores will be calculated by summation of the responses of items shown in Table 3, and divided by the standard deviation of siblings.

Sample weighting: Both the NHIS and BRFSS incorporate intentional oversampling techniques to target certain demographic characteristics and accurately represent the U.S. population.^{32,33} Each participant is given a weighting value which determines the relative contribution to each survey result to create an estimate of the U.S. population. However, the CCSS population differs from the U.S. general population based on key demographics, so direct comparison between the CCSS and national surveys is challenging. Therefore, to compare responses from FU-6 to similar items from the national surveys, person weights from the NHIS/BRFSS samples will be accounted for in multivariable models, to match the composition of the CCSS survivor and sibling samples. The weighting variable in the NHIS to be used is PERWEIGHT; the weighting variable for the BRFSS is _LLCPWT. Adjustments will be race, ethnicity, sex, age, educational attainment, marital status, and presence of chronic medical conditions.

Demographic analysis: The initial analysis will gather frequencies, means/SDs, and medians/IQRs of demographic, disease, and treatment-related information to describe siblings of childhood cancer survivors as well as the general population cohorts. This information will be summarized in the demographic table (Table 5) below.

Aim 1: Describe the prevalence of financial hardship in siblings of childhood cancer survivors. The proportion of positive responses to each individual item pertaining to financial hardship will be reported according to Table 6 below.

Aim 2: Compare the prevalence of financial hardship in siblings of childhood cancer survivors with adults from the general population without a cancer history.

The proportion of positive responses from siblings to each individual item of interest in FU-6 and the corresponding national surveys will be compared. Multivariable weighted logistic regression models pertaining to each individual item will be calculated to compare CCSS siblings to adults from the general population, with sample weights as above. Results will be summarized in Table 7.

Aim 3: Identify characteristics associated with greater financial hardship in siblings of cancer survivors.

For each financial hardship domain as listed in Table 3, we will evaluate the following sibling demographic characteristics for associations with greater risk in siblings: age, sex, race, ethnicity, educational attainment, marital status, household income, chronic health conditions, insurance status, debt and assets, and out-of-pocket health spending. Some additional questions that were not used in domain score creation may also be analyzed. Relative differences for each association would be reported as odds ratios (ORs) as in Table 8. Similarly, for each item we will evaluate associations based on the following survivor disease and treatment-related characteristics (Table 4): type of cancer, age at diagnosis, time from diagnosis, sex, marital status, chronic conditions, receipt of radiation, surgery (without chemotherapy/radiation), transplant status, and history of late relapse. Relative differences for each association would be reported as ORs shown in Table 9.

Table 5: Sociodemographic characteristics of CCSS and national survey respondents.

Characteristic	CCSS Siblings (n=) N %	NHIS Respondents (n=) N %	BRFSS Respondents (n=) N %
Age at questionnaire			
Sex Female Male Other			
Race and ethnicity Black, Non-Hispanic White, Non-Hispanic Hispanic/Latin Other			
Chronic health conditions (%)			
Household income (\$) <20,000 20,000-59,999 60,000-99,999 ≥100,000			
Education <High school High school-some college College graduate or greater			
Marital status Married Singled Divorced or separated Widowed			

Table 6: Prevalence of individual items of financial hardship experienced by siblings.

FU-6 Item	Question	Prevalence (%)
C4	How concerned are you that you may not be able to maintain your current level of insurance coverage over the next 12 months?	
C5	In the past 12 months, did you/anyone in the family have problems paying or were unable to pay any medical bills? Include bills for doctors, dentists, hospitals, therapists, medication, equipment, nursing home or home?	
C6	Do you or anyone in your family currently have medical bills that you are unable to pay at all?	
C7	Do you or anyone in the house you live in have medical bills that are being paid off over time?	
C8	During the past 12 months, was there a time when you needed one of the following, but did not get it because you couldn't afford it? Check all that apply.	
C8a	Any needed medical care	
C8b	Yearly visit to your PCP	
C8c	Prescription medicine	
C8d	Mental health care or counseling	
C8e	Dental care	
C8f	Eye care	
C8g	Care from a specialist	

C9	Have you or your family had to make any other kinds of financial sacrifices in the past 2 years because of debt related to medical care? Check all that apply.	
C9a	Reduced spending on vacation or leisure activities	
C9b	Reduced spending on purchasing large items (e.g. a car)	
C9c	Reduced spending on basics (e.g. food and clothing)	
C9d	Delayed or reduced spending on home improvement	
C9e	Used savings set aside for other purposes (e.g. retirement, educational funds, family support)	
C9f	Made a change to living situation (e.g. sold, refinanced or moved to a smaller residence).	
C10	During the past year, about how much did you spend out-of-pocket for your medical care?	
C11	Has your physical condition or medical treatment caused you financial difficulties over the past week?	
C12	Has your physical condition or medical treatment caused you financial difficulties over the past year?	
C13	In the past 2 years, were you working for pay either full time or part time at a job or business?	
C14	In the past 2 years, did you ever:	
C14a	Take extended paid time off from work, unpaid time off, or make a change in your hours, duties or employment status?	
C14b	Change to a less demanding job?	
C14c	Change from a set work schedule, where you start and end at the same the time every day, to a flexible work schedule, where your start and end times vary from day to day?	
C14d	Decide not to pursue an advancement or promotion?	
C14e	Worry that you might be forced to retire or quit work before you are ready?	
C14f	Stay at a job in part because you were concerned about losing your health insurance?	
C15	Did your spouse or significant other ever stay at a job in part because he/she was concerned about losing health insurance for the family?	
C16	If you get sick or have an accident, how worried are you that you will not be able to pay your medical bills?	
C17	How often in the last 12 months would you say you were worried or stressed about having enough money to pay your rent or mortgage?	
C18	How often in the last 12 months would you say you were worried or stressed about having enough money to buy nutritious meals?	
C19	How often in the last 12 months would you say you were worried or stressed about having enough money to pay household utilities, including water, gas, and electricity?	
C29	Have you ever been sent to collections because of debts you were unable to pay on time or at all?	
C30	Have you ever filed for bankruptcy because of debts you were unable to pay?	

Table 7: Financial hardship among siblings of cancer survivors and the general population.

	CCSS Siblings (n=)	General Population (n=)
	N %	N %
Questions related to difficulty affording necessities		
Problems paying medical bills (C5)		
Unable to pay medical bills (C6)		
Difficulty affording food (C18)‡		
Rent, mortgage, or utilities insecurity (C17/19)‡		

Questions related to coping behaviors Foregoing any medical care (C8a) Prescription medications (C8b) Mental health care (C8c) Dental care (C8d)		
Question related to psychological worry Worried about ability to pay bills if getting sick (C16)		

†Responding to the NHIS

‡Responding to the BRFSS

Table 8: Associations of sibling characteristics and financial hardship domain scores.

Hardship category	Behavioral hardship OR (95% CI)	Material hardship OR (95% CI)	Psychological hardship OR (95% CI)	Additional items in additional columns
Age at questionnaire				
Sex Female (ref) Male Other				
Race and ethnicity Black, Non-Hispanic White, Non-Hispanic (ref) Hispanic/Latin Other				
Education <High school High school-some college College graduate or greater (ref)				
Marital status Married (ref) Singled Divorced or separated Widowed				
Household income (\$) <20,000 20,000-59,999 60,000-99,999 ≥100,000 (ref)				
Chronic health conditions None (ref) 1 condition >1 condition				
Insurance coverage None Individual or employer-based (ref) Medicare Medicaid or IHS Other				
Insurance deductible Less than \$1,300 (ref) \$1,301-2600 Greater than \$2,600				
Out-of-pocket expenses <10% of annual income (ref) ≥10% of annual income				
Homeownership Yes (ref)				

No				
Home value \$0-\$100,000 \$100,001-\$250,000 \$250,001-\$500,000 \$500,001 or more (ref)				
Debt on home mortgage None (ref) \$0-\$50,000 \$50,001-\$100,000 \$100,001-\$250,000 \$250,001-\$500,000 \$500,001 or more				
Other debts None (ref) \$0-\$25,000 \$25,001-50,000 \$50,001-\$100,000 \$100,001-\$250,000 \$250,001-\$500,000 \$500,001 or more				

Table 9: Associations of disease and treatment-related factors with financial hardship of siblings.

	Item C1 Siblings OR (95% CI)	Item C3 Siblings OR (95% CI)	Additional items in additional columns
Age at diagnosis 0-4.99 years 5-9.99 years 10-14.99 years 15-20 years			
Time elapsed from diagnosis 10-19.99 years 20-29.99 years 30-39.99 ≥40 years			
Sex Female Male			
Diagnosis Leukemia CNS tumor Hodgkin Lymphoma Non-Hodgkin Lymphoma Wilms tumor Neuroblastoma Soft tissue sarcoma Bone cancer			
Chronic health conditions None 1 condition >1 condition			
Marital status Married Singled Divorced or separated Widowed			
Radiation			

None TBI only Cranial RT, no TBI Chest RT without Cranial TBI Other RT			
Surgery only (no chemotherapy/radiation) Yes No			
Stem cell transplant Yes No			
History of late relapse Yes No			

LIMITATIONS

- While this proposal uses individuals from the general population as the control cohort, rather than siblings, the sociodemographic composition of the control groups was purposely oversampled and will likely differ significantly from the survivor and sibling cohorts, which is a largely white population. The proposed sample weighting approach will be used to compensate for this.
- The BRFSS Social Determinants of Health special section was only administered in 17 states, which may limit generalizability of the questions matched to the BRFSS. Additionally, the question from BRFSS relating to material hardship is not identical to the questions on FU-6. A composite measure of FU-6 questions will need to be created to compare to the BRFSS, potentially leading to differential misclassification and bias of the data toward or away from the null hypothesis.
- Many question items from the FU-6 form did not have a matching correlate to these national surveys for comparison. Thus, only within-group comparisons can be made for such items, rather than domain scores as proposed in Aim 3.

REFERENCES

1. Robison LL, Hudson MM. Survivors of childhood and adolescent cancer: life-long risks and responsibilities. *Nat Rev Cancer*. 2014;14(1):61-70. doi:10.1038/nrc3634
2. Nathan PC, Nachman A, Sutradhar R, et al. Adverse mental health outcomes in a population-based cohort of survivors of childhood cancer: Mental Health Outcomes in CCS. *Cancer*. 2018;124(9):2045-2057. doi:10.1002/cncr.31279
3. Hudson MM, Ness KK, Gurney JG, et al. Clinical Ascertainment of Health Outcomes Among Adults Treated for Childhood Cancer. *JAMA*. 2013;309(22):2371. doi:10.1001/jama.2013.6296
4. Dowling E, Yabroff KR, Mariotto A, McNeel T, Zeruto C, Buckman D. Burden of illness in adult survivors of childhood cancers: Findings from a population-based national sample. *Cancer*. 2010;116(15):3712-3721. doi:10.1002/cncr.25141

5. Altice CK, Banegas MP, Tucker-Seeley RD, Yabroff KR. Financial Hardships Experienced by Cancer Survivors: A Systematic Review. *JNCI J Natl Cancer Inst.* 2017;109(2):djw205. doi:10.1093/jnci/djw205
6. Nathan PC, Henderson TO, Kirchhoff AC, Park ER, Yabroff KR. Financial Hardship and the Economic Effect of Childhood Cancer Survivorship. *JCO.* 2018;36(21):2198-2205. doi:10.1200/JCO.2017.76.4431
7. Parsons SK, Castellino SM, Yabroff KR. Cost, Value, and Financial Hardship in Cancer Care: Implications for Pediatric Oncology. *American Society of Clinical Oncology Educational Book.* 2018;(38):850-860. doi:10.1200/EDBK_200359
8. Guy GP, Yabroff KR, Ekwueme DU, Rim SH, Li R, Richardson LC. Economic Burden of Chronic Conditions Among Survivors of Cancer in the United States. *JCO.* 2017;35(18):2053-2061. doi:10.1200/JCO.2016.71.9716
9. Kirchhoff AC, Krull KR, Ness KK, et al. Occupational outcomes of adult childhood cancer survivors: A report from the childhood cancer survivor study. *Cancer.* 2011;117(13):3033-3044. doi:10.1002/cncr.25867
10. Guy GP, Berkowitz Z, Ekwueme DU, Rim SH, Yabroff KR. Annual Economic Burden of Productivity Losses Among Adult Survivors of Childhood Cancers. *PEDIATRICS.* 2016;138(Supplement):S15-S21. doi:10.1542/peds.2015-4268D
11. Nipp RD, Kirchhoff AC, Fair D, et al. Financial Burden in Survivors of Childhood Cancer: A Report From the Childhood Cancer Survivor Study. *JCO.* 2017;35(30):3474-3481. doi:10.1200/JCO.2016.71.7066
12. Huang IC, Bhakta N, Brinkman TM, et al. Determinants and Consequences of Financial Hardship Among Adult Survivors of Childhood Cancer: A Report From the St. Jude Lifetime Cohort Study. *JNCI: Journal of the National Cancer Institute.* 2019;111(2):189-200. doi:10.1093/jnci/djy120
13. Kale HP, Carroll NV. Self-reported financial burden of cancer care and its effect on physical and mental health-related quality of life among US cancer survivors. *Cancer.* 2016;122(8):283-289. doi:10.1002/cncr.29808
14. Ramsey S, Blough D, Kirchhoff A, et al. Washington State cancer patients found to be at greater risk for bankruptcy than people without a cancer diagnosis. *Health Aff (Millwood).* 2013;32(6):1143-1152. doi:10.1377/hlthaff.2012.1263
15. Fair D, Park ER, Nipp RD, et al. Material, behavioral, and psychological financial hardship among survivors of childhood cancer in the Childhood Cancer Survivor Study. *Cancer.* 2021;127(17):3214-3222. doi:10.1002/cncr.33613
16. Nathan PC, Huang IC, Chen Y, et al. Experiences of financial hardship in adult survivors of childhood cancer: A report from the Childhood Cancer Survivor Study (CCSS). Published online 2021.

17. Zheng Z, Han X, Zhao J, et al. Financial Hardship, Healthcare Utilization, and Health Among U.S. Cancer Survivors. *American Journal of Preventive Medicine*. 2020;59(1):68-78. doi:10.1016/j.amepre.2020.02.016
18. Coughlin SS, Datta B, Berman A, Hatzigeorgiou C. A cross-sectional study of financial distress in persons with multimorbidity. *Preventive Medicine Reports*. 2021;23:101464. doi:10.1016/j.pmedr.2021.101464
19. Yabroff KR, Dowling E, Rodriguez J, et al. The Medical Expenditure Panel Survey (MEPS) Experiences with Cancer Survivorship Supplement. *J Cancer Surviv*. 2012;6(4):407-419. doi:10.1007/s11764-012-0221-2
20. Gerhardt CA, Lehmann V, Long KA, Alderfer MA. Supporting Siblings as a Standard of Care in Pediatric Oncology: Standard of Care for Siblings. *Pediatr Blood Cancer*. 2015;62(S5):S750-S804. doi:10.1002/pbc.25821
21. Erker C, Yan K, Zhang L, Bingen K, Flynn KE, Panepinto J. Impact of pediatric cancer on family relationships. *Cancer Med*. 2018;7(5):1680-1688. doi:10.1002/cam4.1393
22. Morales S, Salehabadi SM, Srivastava D, et al. Health-related and cancer risk concerns among siblings of childhood cancer survivors: a report from the Childhood Cancer Survivor Study (CCSS). *J Cancer Surviv*. Published online June 1, 2021. doi:10.1007/s11764-021-01056-0
23. Buchbinder D, Casillas J, Krull KR, et al. Psychological outcomes of siblings of cancer survivors: a report from the Childhood Cancer Survivor Study. *Psychooncology*. 2011;20(12):1259-1268. doi:10.1002/pon.1848
24. Houtzager BA, Grootenhuis MA, Hoekstra-Weebers JEHM, Caron HN, Last BF. Psychosocial functioning in siblings of paediatric cancer patients one to six months after diagnosis. *European Journal of Cancer*. 2003;39(10):1423-1432. doi:10.1016/S0959-8049(03)00275-2
25. Houtzager BA, Grootenhuis MA, Caron HN, Last BF. Quality of life and psychological adaptation in siblings of paediatric cancer patients, 2 years after diagnosis. *Psycho-Oncology*. 2004;13(8):499-511. doi:10.1002/pon.759
26. van Warmerdam J, Sutradhar R, Kurdyak P, et al. Long-Term Mental Health Outcomes in Mothers and Siblings of Children With Cancer: A Population-Based, Matched Cohort Study. *JCO*. 2020;38(1):51-62. doi:10.1200/JCO.19.01382
27. Lund LW, Winther JF, Dalton SO, et al. Hospital contact for mental disorders in survivors of childhood cancer and their siblings in Denmark: a population-based cohort study. *The Lancet Oncology*. 2013;14(10):971-980. doi:10.1016/S1470-2045(13)70351-6
28. Kim Y, Given BA. Quality of life of family caregivers of cancer survivors: Across the trajectory of the illness. *Cancer*. 2008;112(S11):2556-2568. doi:10.1002/cncr.23449
29. Robison LL, Mertens AC, Boice JD, et al. Study design and cohort characteristics of the Childhood Cancer Survivor Study: a multi-institutional collaborative project. *Med Pediatr Oncol*. 2002;38(4):229-239. doi:10.1002/mpo.1316

30. Robison LL, Armstrong GT, Boice JD, et al. The Childhood Cancer Survivor Study: a National Cancer Institute-supported resource for outcome and intervention research. *J Clin Oncol.* 2009;27(14):2308-2318. doi:10.1200/JCO.2009.22.3339
31. National Center for Health Statistics. *Survey Description, National Health Interview Survey, 2019.*; 2020. Accessed October 16, 2021. https://ftp.cdc.gov/pub/Health_Statistics/NCHS/Dataset_Documentation/NHIS/2019/srvydesc-508.pdf
32. Parsons VL, Moriarity C, Jonas K, Moore TF, Davis KE, Tompkins L. Design and estimation for the national health interview survey, 2006-2015. *Vital Health Stat 2.* 2014;(165):1-53.
33. Center for Disease Control. The Behavioral Risk Factor Surveillance System: Complex Sampling Weights and Preparing 2017 BRFSS Module Data for Analysis. Published online July 2018. Accessed October 21, 2021. https://www.cdc.gov/brfss/annual_data/2017/pdf/Complex-Smple-Weights-Prep-Module-Data-Analysis-2017-508.pdf