

Financial Hardship in Survivors of Childhood Cancer and their Siblings: Overview paper

Working Group: Cancer control

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Background

The impact of the diagnosis and treatment of childhood or adolescent cancer on long-term physical and psychological health of survivors has been described extensively. Less well understood is the financial hardship that can arise from childhood cancer, for both the survivor and their family members. Physical, neurocognitive and psychological late effects may lead to financial hardship and in turn, financial hardship may exacerbate or cause physical and psychological harms. Indeed, data from the National Health Interview Survey (NHIS) have shown that in the general population, financial hardship is correlated with poor physical and mental health, reduced satisfaction with social activities and relationships, and poorer quality of life.¹

Among adults with a current or prior cancer, studies have demonstrated that individuals with cancer face more financial hardship than those without a cancer history.²⁻⁵ Adult cancer survivors have higher out-of-pocket medical costs, in part due to late effects of their cancer therapy,⁵⁻⁷ and are more likely to be unable to work because of poor health and functional limitations.⁵⁻⁷ This may limit their access to employment-based health insurance, their income and asset accumulation and, consequently, their ability to pay for needed health care.⁸

Such financial hardship has been demonstrated to manifest itself among adult cancer patients and survivors in several ways, including:

- High out-of-pocket medical costs that impact household income and asset accumulation
- Inability to pay for medical care or deferral of needed care or testing
- Asset depletion, general/medical debt or declaration of bankruptcy
- Housing and food insecurity

- Elevated stress, distress or worry about current/future financial situation
- Inability to actively engage in productive work for pay due to lasting effects of cancer, causing cancer survivors and/or their caregivers and families to be unemployed, underemployed, uninsured or underinsured
- Job lock, or the inability of individuals to freely engage in extra work activities to earn a promotion, reduce work hours to attend to household needs including but not limited to health needs, or to leave a job to pursue other activities because doing so will result in the loss of employee benefits, most notably health insurance

Medical financial hardship can be broadly categorized into three domains: *material*, *psychological*, and *behavioral*. Material hardship is typically measured by objective factors such as medical debt, out-of-pocket expenses for medical costs, or difficulty paying medical bills. Psychological hardship is the stress, distress, and worry that occurs over a current or future financial situation related to medical needs. Behavioral hardship captures actions such as delaying or forgoing medical care, or treatment non-adherence due to concerns over costs. All domains contribute to the full burden of financial hardship that can impact survivors of cancer.

Only a handful of studies in the child and young adult cancer population have examined aspects of financial hardship. Among adults with cancer, several studies have demonstrated an association between younger age at initial cancer diagnosis and risk for financial hardship^{12,13} such as higher rates of debt and bankruptcies.⁹ Lack of savings, competing financial obligations (e.g. children) and absence of access to comprehensive insurance coverage including catastrophic limits on out of pocket spending are particular challenges in this demographic.¹⁰ Whether similar factors contribute to hardship in adult survivors of childhood cancers is less well established. Using items measuring ability to pay monthly bills and financial worries, the St. Jude Lifetime Cohort Study (SJLIFE) identified that 49%, 37%, and 14% of adult survivors of childhood cancer experienced low, moderate, and high level of financial hardship, respectively.

CCSS has previously investigated some elements of financial hardship in a sub-set of survivors in the original (1970-86) cohort, providing proof of concept for this proposal. Elyse Park and colleagues conducted an ancillary study entitled: “*Are adult survivors of childhood cancer underinsured?*” This study generated several publications addressing insurance and underinsurance in a randomly chosen subset of 698 survivors and 210 siblings, as well as a sample of 39 survivors who underwent in-depth interviews.¹¹⁻¹⁴ Almost one quarter of these survivors had skipped or deferred medical care or testing or have had difficulty paying medical bills, and 10% reported substantial out-of-pocket medical costs (>10% of their income).¹⁵

The CCSS follow-up 6 survey focused specifically on financial hardship. The conceptual framework of the survey was consistent with economic models of financial hardship and its moderators among adults with cancer and survivors published by the NCI (www.cancer.gov/about-cancer/managing-care/financial-toxicity-hp-pdq). The questions were consistent with recommendations from the NCI PDQ working group regarding future studies of

financial hardship. The questions were derived from population-based nationally representative surveys that have undergone extensive cognitive testing. This will allow for comparison to general population data in future analyses. These surveys were: National Health Interview Survey (NHIS; www.cdc.gov/nchs/nhis/); Medical Expenditure Panel Survey (MEPS; www.healthcaresdelivery.cancer.gov/meps) and the Behavioral Risk Factor Surveillance System (BRFSS; www.cdc.gov/brfss/).

This CCSS survey was sent to a subset of the survivor and sibling cohorts. Overall, 4151 survivors and 1039 siblings responded. Attached is the first of three concept proposals that will form the preliminary analyses of the financial hardship data. This overview paper will examine: assets/debt, financial hardship related to medical care, and food/shelter insecurity. The other two analyses (to be submitted separately) will: 1) evaluate the relationship between chronic health conditions and financial hardship; and 2) examine the relationship between health insurance, employment and financial hardship, including the concept of “job lock”, which is when a subject reports being ‘locked’ in their present employment or health insurance. Several subsequent analyses (including comparison to general population data from NHIS, MEPS and BRFSS, and a separate analysis focused on sibling outcomes) are planned.

Hypotheses

- 1) We hypothesize that survivors will report medical and non-medical financial hardship more often than their siblings, as manifested by:
 - a) *Medical financial hardship*
 - i) *Material*: High out of pocket medical expenses (>10% of family income), history of being sent to debt collection, history of filing for bankruptcy, problems paying off medical bills, need to pay medical bills off over time, inability to afford needed medical care
 - ii) *Psychological*: Worry about being able to afford medical costs if they become ill or are involved in an accident
 - iii) *Behavioral*: Delaying of foregoing medical care (including outpatient care, prescription medication, mental health care, dental care, eyeglasses, specialist or survivor follow-up care because of cost)
 - b) *Non-medical financial hardship: food and housing insecurity*
- 2) Among survivors, a higher intensity of financial hardship (i.e. endorsement of more sub-categories with each of material, psychological and behavioral hardship; and a higher likelihood of manifesting at least one measure of hardship within each category) will be associated with:
 - a) Clinical characteristics related to primary cancer diagnosis and treatment
 - i) Primary cancer diagnoses associated with high burden of late effects (e.g. CNS tumors, Hodgkin lymphoma)

- ii) Higher doses of anthracycline or alkylating chemotherapy
 - iii) Receipt of radiation (particularly cranial or chest)
 - iv) Stem cell transplant
 - v) Shorter time since last treatment
- b) Sociodemographic characteristics
- i) Female gender
 - ii) Minority race/ethnicity
 - iii) Younger age at survey
 - iv) Younger age at diagnosis
 - v) Presence and type of health insurance coverage
 - vi) Household assets (owning home, home value)
 - vii) Local median income/unemployment rate (based on zip code at time of survey)

Methods/Analysis

Subject population:

The study sample will consist of all survivors and siblings who were sent and responded to the Financial Hardship question as part of F/U #6.

Outcomes of interest:

We will focus on selected questions from the follow up #6 survey (see Appendix).

Hypothesis 1 Outcomes:

- 1) High out of pocket medical spending (>10% of family income) (C10, C21)
- 2) History of being sent to debt collection (C29)
- 3) History of filing for bankruptcy (C30, C31)
- 4) Problems paying medical bills (C5, C6)
- 5) Inability to afford medical/paramedical care or delay/foregoing of medical care (C8)
- 6) Worries about not being able to pay upcoming medical bills (C16)
- 7) Concern about being able to pay mortgage, afford food, or pay for household utilities (C17, C18, C19)
- 8) Financial sacrifices because of medical debt (C9)

Hypothesis 2 Outcomes:

For analyses among survivors, we will measure medical financial hardship intensity characterized by the number (0, 1, 2 or 3) of financial hardship categories (material,

psychological, behavioral) in which they report at least one measure of hardship (Method A). Although there are not the same number of items in each of the three categories, this approach to estimating intensity has been used in several prior analyses of financial hardship. In addition to counting the total number of hardship categories (0-3), we will consider statistical approaches to measuring financial hardship intensity, such as factor analysis to generate hardship intensity scores for individual hardship categories and across three categories (Method B). Factor analysis allows for incorporating relative weights of individual hardship items (i.e., factor loadings) into the calculation of hardship intensity scores. First, we will perform principal factor analysis with the multiple items of each hardship category to generate a factor score for each participant. Specific items with low factor loadings (e.g., standardized loading <0.3) may be removed from the score calculation after reviewing the appropriateness of content. Second, the standardized factor scores derived from each of three categories will be summed and then trichotomized by Z-scores <-1 , -1 to 1 , and >1 (or per score distribution across all study participants) for measuring the overall hardship intensity (as low, moderate, high level).

Candidate variables to be considered in assessing these outcomes will include (see tables for initial categorizations):

- Sociodemographic variables:
 - Age
 - Gender
 - Race/ethnicity
 - Highest education level
 - Current employment status
 - Insurance coverage
 - Household income
 - Marital status
- Disease/treatment variables:
 - Cancer diagnosis
 - Age at diagnosis
 - Doxorubicin equivalent dose
 - Cyclophosphamide equivalent dose
 - If yes to radiation → cranial radiation, mantle/chest radiation, TBI

Not all of the socioeconomic variables can be included together in multivariable analyses because of collinearity. The current analysis will not examine the relationship between chronic health conditions or health status variables and financial outcomes since these will be the focus of subsequent papers.

Data analysis plan

Initial analyses will focus on generating frequencies for categorical variables and mean/SD or median/IQR for continuous variables describing characteristics of the survivor and sibling cohorts (Table 1). Hypothesis 1 analyses will utilize binary outcomes defined above and comparisons between survivors and siblings will be carried out using generalized linear models (logistic or log link functions, depending on prevalence) to estimate relative rates between groups, adjusted for age, sex, race/ethnicity and marital status. Measures of socioeconomic status will be assessed separately for different outcome measures. For example, household income will not be included in the evaluation of out-of-pocket spending as a percentage of family income, but will be included in the evaluation of outcomes related to affordability.

Among survivors, in analyses for Hypothesis 2, after generating the outcome measures as described above using Methods A and B, we will fit univariate and multivariable ordinal regression models to each of the ordered categorical outcomes to evaluate associations of sociodemographic, diagnostic and treatment variables with medical financial hardship intensity outcome variables defined above. *A priori*, we plan to adjust all models for age and sex. Factors that are significant at the $p < 0.1$ level in adjusted univariate analyses will be included in multivariable analyses as candidates for inclusion, taking into account which variables are collinear and choosing between them based on importance in the model. Step-down model fitting will be carried out using model fit metrics such as QIC as well as statistical significance.

Examples of tables and figures

Table 1: Characteristics of adult survivors of childhood cancer and siblings

Characteristic:	Survivors (n=)		Siblings (n=)	
	n	%	n	%
Sex				
Male				
Female				
Age at diagnosis (median, range)				
Race/ethnicity				
White, Non-Hispanic				
Black, Non-Hispanic				
Hispanic/Latin				
Other				
Age at questionnaire				
18-24 years				
25-34 years				
35-39 years				
40-44 years				
≥45 years				
Education*				
<College				

College graduate

Employment*

Employed or caring for home
Looking for work or unable to work

Household income (\$)*

<20,000
20,000-59,999
60,000-99,999
≥100,000

Insurance Coverage*

Private only
Any public
Uninsured

Marital Status*

Married
Single
Divorced or separated

Age at diagnosis

0-4 years
5-9 years
10-14 years
15-20 years

Cancer Diagnosis

Leukemia
Lymphoma
CNS tumor
Kidney tumor
Neuroblastoma
Bone/Soft tissue sarcoma

**Anthracycline Chemotherapy
(doxorubicin equivalent dose)**

None
< 250 mg/m²
≥ 250 mg/m²

Alkylating agent Chemotherapy (cyclophosphamide equivalent dose)

None
< 4000 mg/m²
4000-8000 mg/m²
≥8000 mg/m²

H SCT

Yes
No

Radiation

Cranial
Chest
TBI
Other
None

Table 2: Medical Financial Hardship in Adult Survivors of Childhood Cancer and Siblings

	Cancer Survivors (n =)		Siblings (n =)		<i>P</i>
	N	%	N	%	
<i>Material</i>					
Out-of-pocket medical spending > 10% of family income					
Sent to debt collection					
(Debt – assets) > xx% of family income					
Ever filed for bankruptcy protection					
Problems paying medical bills					
Paying off medical bills over time					
Could not afford any needed medical care					
Any material hardship					
<i>Psychological</i>					
Worried about medical costs of illness/accident					
Worried about medical costs of healthcare					
Any psychological hardship					
<i>Behavioral</i>					
Delayed any medical care due to cost					
Forgone any medical care due to cost					
Delayed or forgone					
prescription medicine					
mental health care/counselling					
dental care					
eyeglasses					
specialist					
follow-up care					
Any behavioral hardship					

Figure 1: Figure with frequency of financial sacrifices – reduced spending on vacation, purchasing large items, spending on basics, using savings, change to living situation for survivors and siblings

Figure 2: Figure with non-medical financial hardship worry about paying rent/mortgage, worry about buying meals, worry about paying utilities for survivors and siblings (always, usually, sometimes vs rarely/never, DK/UK)

Table 3. Associations of Cancer Survivor Characteristics and Medical Financial Hardship

Intensity

<u>Characteristic:</u>	Financial Hardship Intensity Unadjusted		Financial Hardship Intensity Unadjusted	
	Moderate intensity ^{\$,†}	High intensity ^{\$,‡}	Moderate intensity ^{\$,†}	High intensity ^{\$,‡}
	OR	95% CI	OR	95% CI
Sex				
Male				
Female				
Race/ethnicity				
White, Non-Hispanic				
Black, Non-Hispanic				
Hispanic/Latin				
Other				
Age at questionnaire				
18-24 years				
25-34 years				
35-39 years				
40-44 years				
≥45 years				
Education*				
<College				
College graduate				
Employment*				
Employed or caring for home				
Looking for work or unable to work				
Household income (\$)*				
<20,000				
20,000-59,999				
60,000-99,999				
≥100,000				

Insurance Coverage*

Private only
Any public
Uninsured

Marital Status*

Married
Single
Divorced or separated

Age at diagnosis

0-4 years
5-9 years
10-14 years
15-20 years

Cancer Diagnosis

Leukemia
Lymphoma
CNS tumor
Kidney tumor
Neuroblastoma
Bone/Soft tissue sarcoma

**Anthracycline Chemotherapy
(doxorubicin equivalent dose)**

None
< 250 mg/m²
≥ 250 mg/m²

Alkylating agent Chemotherapy (cyclophosphamide equivalent dose)

None
< 4000 mg/m²
4000-8000 mg/m²
≥8000 mg/m²

HSCT

Yes
No

Radiation

Cranial
Chest
Other
None

§ Low intensity (as the reference group): defined as number of hardship categories = 0 (Model A) and hardship intensity Z-scores <-1 (Model B)

† Moderate intensity: defined as number of hardship categories =1 (Model A) and hardship intensity Z-scores -1 to 1 (Model B)

‡ High intensity: defined as number of hardship categories =2 or 3 (Model A) and hardship intensity Z-scores > 1 (Model B)

Appendix: Selected questions from follow-up survey

Medical care (Section C)

5. In the past 12 months, did you or anyone in the house you live in have problems paying or were unable to pay any medical bills? Include bills for doctors, dentists, hospitals, therapists, medication, equipment, nursing homes or home care.
 - Yes
 - No →SKIP TO C7
 - Don't know → SKIP to C7

6. Do you or anyone in the house you live in currently have medical bills that you are unable to pay at all?
 - Yes
 - No

7. Do you or anyone in the house you live in have medical bills that are being paid off over time? This could include medical bills being paid off with a credit card, through personal loans, bill paying arrangements with hospitals or other providers, or collection agencies.
 - Yes
 - No

8. 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.

	Yes	No
Any needed medical care		
Yearly visit to your primary care doctor		
Prescription medicine		
Mental health care or counseling		
Dental care		
Eyeglasses		
Care from a specialist		
Survivor care or screening		

9. Have you or anyone in your house had to make any other kinds of financial sacrifices in the past 5 years because of debt related to medical care? Check all that apply.
 - Reduced spending on vacation or leisure activities
 - Reduced spending on purchasing large items (e.g. a car)
 - Reduced spending on basics (e.g. food and clothing)
 - Delayed or reduced spending on home improvement
 - Used savings set aside for other purposes (e.g. retirement, educational funds, family support)
 - Made a change to living situation (e.g. sold, refinanced or moved to a smaller residence).
 - Other. Please specify: _____

10. During the past year, about how much did you spend out-of-pocket for your medical care? Include out-of-pocket expenses for prescription drugs, co-payments, and deductibles, but do not include health insurance premiums or any costs paid by your health insurance.
 \$ _____

11. Has your physical condition or medical treatment caused you financial difficulties over the past week?
 - No difficulty
 - A little difficulty
 - Quite a bit of difficulty
 - Great difficulty

12. Has your physical condition or medical treatment caused you financial difficulties over the past year?
- No difficulty
 - A little difficulty
 - Quite a bit of difficulty
 - Great difficulty

Other worries (Section C)

16. 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
17. 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
 - Prefer not to say
 -
18. 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
 - Prefer not to say

19. How often in the last 12 months would you say you were worried or stressed about having enough money to pay household utilities such as water, gas, and electricity?
- Always
 - Usually
 - Sometimes
 - Rarely
 - Never
 - Don't know
 - Prefer not to say

Assets and Debt (section C)

22. Does anyone in your immediate family (you, your spouse or significant other, your child, your sibling, or your parent) own the home you are living in?
- Yes
 - No →SKIP TO QUESTION C27
23. Who in your family owns your home? (Check all that apply)
- You
 - Your spouse or significant other
 - Someone else in your family

24. Approximately what is the value of your home if it was sold today?
- \$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
25. Are there any mortgages or other loans outstanding on this home?
- Yes
 - No →SKIP TO QUESTION C27
 - Don't know →SKIP TO QUESTION C27
26. How much is currently owed on these mortgages or loans?
- \$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
27. Do you or anyone in your house have other debts such as credit card balances, car loans, debts owed to medical providers, life insurance policy loans, loans from relatives and so forth?
- Yes
 - No →SKIP TO next section
28. What is the total amount owed on this other debt?
- \$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
29. Have you ever been sent to collections because of debts you were unable to pay on time or at all?
- Yes
 - No
30. Have you ever filed for bankruptcy because of debts you were unable to pay?
- Yes
 - No → SKIP TO next section
31. What was the most recent year in which you filed for bankruptcy? _ _ _ _

References

1. Fenn KM, Evans SB, McCorkle R, et al: Impact of financial burden of cancer on survivors' quality of life. *J Oncol Pract* 10:332-8, 2014
2. Soni A: Trends in the Five Most Costly Conditions among the U.S. Civilian Institutionalized Population, 2002 and 2012. , Statistical Brief 470, Agency for Healthcare Research and Quality., 2015
3. Bradley CJ, Yabroff KR, Warren JL, et al: Trends in the Treatment of Metastatic Colon and Rectal Cancer in Elderly Patients. *Med Care* 54:490-7, 2016
4. Yabroff KR, Dowling EC, Guy GP, Jr., et al: Financial Hardship Associated With Cancer in the United States: Findings From a Population-Based Sample of Adult Cancer Survivors. *J Clin Oncol* 34:259-67, 2016
5. Ekwueme DU, Yabroff KR, Guy GP, Jr., et al: Medical costs and productivity losses of cancer survivors--United States, 2008-2011. *MMWR Morb Mortal Wkly Rep* 63:505-10, 2014
6. Guy GP, Jr., Ekwueme DU, Yabroff KR, et al: Economic burden of cancer survivorship among adults in the United States. *J Clin Oncol* 31:3749-57, 2013
7. Guy GP, Jr., Yabroff KR, Ekwueme DU, et al: Healthcare Expenditure Burden Among Non-elderly Cancer Survivors, 2008-2012. *Am J Prev Med* 49:S489-97, 2015
8. Harila-Saari AH, Paakko EL, Vainionpaa LK, et al: A longitudinal magnetic resonance imaging study of the brain in survivors in childhood acute lymphoblastic leukemia. *Cancer* 83:2608-2617, 1998
9. Banegas MP, Guy GP, Jr., de Moor JS, et al: For Working-Age Cancer Survivors, Medical Debt And Bankruptcy Create Financial Hardships. *Health Aff (Millwood)* 35:54-61, 2016
10. PDQ Financial Toxicity and Cancer Treatment. Bethesda, MD, National Cancer Institute, 2016
11. Kirchhoff AC, Parsons HM, Kuhlthau KA, et al: Supplemental security income and social security disability insurance coverage among long-term childhood cancer survivors. *J Natl Cancer Inst* 107:djv057, 2015
12. Kirchhoff AC, Kuhlthau K, Pajolek H, et al: Employer-sponsored health insurance coverage limitations: results from the Childhood Cancer Survivor Study. *Support Care Cancer* 21:377-83, 2013
13. Park ER, Kirchhoff AC, Zallen JP, et al: Childhood Cancer Survivor Study participants' perceptions and knowledge of health insurance coverage: implications for the Affordable Care Act. *J Cancer Surviv* 6:251-9, 2012

14. Park ER, Kirchoff AC, Perez GK, et al: Childhood Cancer Survivor Study Participants' Perceptions and Understanding of the Affordable Care Act. *J Clin Oncol*, 2015

15. Nipp RD, Kirchoff AC, Fair D, et al: Financial Burden in Survivors of Childhood Cancer: A Report From the Childhood Cancer Survivor Study. *J Clin Oncol* 35:3474-3481, 2017