

STUDY TITLE: Continuity and Coordination of Care for Childhood Cancer Survivors with Chronic Conditions (The “7C” Study)

WORKING GROUP: Cancer Control and Intervention

PRINCIPAL INVESTIGATOR: Dr. Claire Snyder, Johns Hopkins University

BACKGROUND

There is an increasing number of survivors of pediatric and adolescent cancer (hereinafter referred to as “childhood cancer survivors”) as a result of more effective therapies,¹ better risk stratification,² and improvements in supportive care. Unfortunately, many of these survivors experience life-long, chronic morbidity associated with their cancer and its treatment.³⁻⁷ By age 50, childhood cancer survivors have an average of 17 chronic health conditions (5 serious/disabling or life-threatening), compared to community controls who have an average of 9 chronic health conditions (2 serious/disabling or life-threatening).⁴ The Institute of Medicine recommends life-long risk-based follow-up of childhood cancer survivors.⁸ But the appropriate roles for primary care providers (PCPs), oncologists, and subspecialists in providing care to childhood cancer survivors with chronic conditions are unclear. Although subspecialists treat many of the morbidities experienced by childhood cancer survivors in patients in the general population, they may have limited knowledge of the pathogenesis, natural history, and context of these conditions in childhood cancer survivors. In addition, given that disparities are prevalent across cancer care, disparities for the particularly vulnerable population of childhood cancer survivors with chronic conditions are likely – but the potential sociodemographic characteristics (e.g., race/ethnicity, income, education, insurance status) that have the greatest influence on the care of childhood cancer survivors is unknown. **Therefore, we propose to conduct a prospective, mixed-methods, longitudinal cohort study with two overarching goals: (1) to examine which provider specialties manage cancer survivors with chronic conditions and how this relates to coordination and continuity of care; and (2) to identify the extent to which various socio-demographic factors are associated with disparities in coordination and continuity of care.** The findings of this study will inform the development of intervention(s) to improve care for this vulnerable population.

The Childhood Cancer Survivor Study (CCSS) is a National Cancer Institute-funded cohort of over 25,000 cancer survivors who were diagnosed before age 21 with one of a number of cancer types between 1970-1999 and who survived at least 5 years (U24 CA55727).⁹ Participants were recruited from 31 centers across the United States and Canada. The study has captured detailed data on cancer diagnosis and treatment and has surveyed the cohort approximately every 2 years to capture a wide range of outcomes, including the burden of morbidity and chronic disease in this population as they age.^{5,7,10-11} Based on the 2014 survey, the most recent one to assess chronic conditions, 4078 CCSS participants report at least 1 severe, life-threatening, or disabling chronic condition. However, the CCSS has historically only collected basic information regarding the health service use of participants, limiting opportunities to study how childhood cancer survivors’ chronic conditions are managed. **By accessing this largest cohort of childhood cancer survivors in North America, this study provides a unique opportunity to address the critical unanswered questions regarding how their chronic conditions are currently being managed, as well as disparities in this management. More importantly, this study will provide insights regarding how this growing population of cancer survivors should be managed to best promote their life-long health and well-being.**

AIMS AND HYPOTHESES

We propose to conduct a prospective, mixed-methods, longitudinal cohort study of treatment patterns and experiences in a sample of CCSS participants with 2 or more chronic conditions (at least 1 of which is severe or disabling/life-threatening). To investigate disparities in care, we will explore the extent to which socio-demographic factors (race/ethnicity, income, education, insurance, and urban/rural location) are associated with patterns, continuity, and coordination of care. The study has the following specific aims:

1. To describe patterns of physician visits (number of visits, types of provider specialties visited) among childhood cancer survivors with chronic conditions.
 - a. To explore which providers survivors consider to be their “responsible provider” (i.e., responsible for most of their health needs and knows the patient well). Responsible providers will be categorized as specialized survivor clinic (PCP- or oncologist-led), other PCP, other oncologist, other specialist (e.g., cardiologist, endocrinologist), no one.
 - b. To identify the association between survivors’ race/ethnicity, income, education, insurance, and urban/rural location regarding their identification of a “responsible provider.”

Hypothesis 1: Compared to other socio-demographic factors, insurance status will have the greatest

association with having no identified responsible provider.

2. To evaluate coordination and continuity of care for childhood cancer survivors with chronic conditions.
 - a. To determine the association between “responsible provider” category (survivorship clinic, other PCP, other oncologist, other specialist, no one) and the coordination and continuity of survivors’ care.

Hypothesis 2a: Participants who identify a survivorship clinic as their “responsible provider” will have better continuity and coordination of care compared to survivors who identify another category of “responsible provider.”
 - b. To evaluate the associations between coordination and continuity of care and with race/ethnicity, income, education, insurance, and urban/rural location.

Hypothesis 2b: Compared to the other factors, insurance will have the strongest association with coordination and continuity of care.
3. Through qualitative interviews, obtain the perspectives of childhood cancer survivors and their providers regarding appropriate roles and responsibilities for patients, PCPs, oncologists, and other specialists in managing chronic conditions in childhood cancer survivors.
4. To use the results from Aims 1-3 to inform the development of an intervention to improve the coordination and continuity of care for childhood cancer survivors with chronic conditions.

METHODS

Study Design: We propose a prospective, mixed-methods, longitudinal cohort study. A subset of participants from the CCSS with 2 or more chronic conditions (at least 1 of which is severe or disabling/life-threatening), will be recruited and followed for 24 months. We will stratify recruitment based on race/ethnicity, income, education, insurance, and urban/rural location.

During the 24-month observation period, we will collect participants’ reports of the number of visits they had and to which provider specialties (Aim 1). As an innovation in this study, we will invite participants to use the MYLTFU patient portal, which was developed for the CCSS, to report on their health service use in real-time. Alternatively, survivors can track their care using hard-copy calendars and report their health service use to the study team at 6-month intervals, similar to what we and others have done in previous studies.^{12,13} These data will be used to describe the participants’ patterns of care, as well as to calculate a measure of coordination risk and of care continuity (Aim 2). We will also assess coordination and continuity of care using validated patient-reported outcome measures collected via survey at 6-month intervals (Aim 2).

For Aim 3, we will conduct qualitative interviews with a subsample of cancer survivors and their primary care and specialist providers. We will purposively sample survivors by race/ethnicity, income, education, insurance, and urban/rural location to obtain diverse perspectives. Interviews will focus on the roles and responsibilities of various physician specialties with respect to (1) general primary care, (2) survivor care (focused on screening, prevention, etc), and (3) treatment for specific chronic conditions. As a complement to Aim 2, we will also obtain survivors’ qualitative perspectives on the quality of care they have received. In addition, we will recruit and interview these survivors’ PCPs, oncology specialists, and other specialists to obtain their perspectives regarding these three areas of care for that particular cancer survivor, as well as for childhood cancer survivors in general. We will also evaluate specialist providers’ knowledge of the sequelae of childhood cancer and confidence in caring for these survivors.

Finally, using the results from Aims 1-3, in Aim 4, we will develop intervention(s) to improve care for childhood cancer survivors with chronic conditions, with particular attention to addressing factors associated with worse continuity and coordination of care. Expecting that the challenges to continuity and coordination of care will occur at multiple levels, development of our intervention(s) will consider addressing patient, caregiver, provider, healthcare team, clinic, delivery organizations, and community factors.

Study Population: The population for this study is childhood cancer survivors with 2 or more chronic conditions (at least 1 of which is severe or life-threatening/disabling) (Aims 1-3) and their providers (Aim 3). Survivor participants will be recruited from the CCSS cohort based on the chronic conditions reported in their most recent CCSS survey (currently in the field) and stratified by race/ethnicity, income, education, insurance, and urban/rural location. To classify the severity of chronic conditions we will apply the Common Terminology Criteria for Adverse Events (CTCAE)¹⁴ grades to the conditions reported through the CCSS, using the methods of Oeffinger et al.⁷ The CTCAE uses five grades: 1=mild, 2=moderate, 3=severe, 4=life-threatening or disabling, 5= fatal. Oeffinger et al have defined CTCAE grades for 137 chronic health conditions. To be eligible for our study, the CCSS participant has to have 2 or more chronic conditions Grade 2 or higher, with at least 1 condition Grade 3 or 4. Based on our preliminary analysis of the CCSS population, approximately 3300 survivors would be eligible for the study based on comorbidity classification. In addition to the chronic condition eligibility criterion, participants must be alive and reside in the US. Survivors who are deceased, physically or cognitively unable to complete the data collection, or reside in Canada will be excluded.

For Aim 3, we will also recruit the oncology, primary care, and other specialty providers of a subset of survivors. This subset of survivors will be selected based on race/ethnicity, income, education, insurance, and urban/rural location at the end of their 24-month observation period so that we will have complete data on their patterns of care. For each of the survivors selected for the qualitative sub-study, we will identify their responsible provider (if any), plus their primary care provider (if any), their primary oncology specialist (if any), and up to 2 providers in other specialties who have played a major role in the survivor's care over the past 24 months. We will recruit these providers for qualitative interviews to obtain their perspectives on the care of the specific cancer survivor participant, as well as care for childhood cancer survivors generally.

Outcome Measures: This study evaluates the quality of comorbid condition care for childhood cancer survivors using patient-centered outcomes that apply regardless of the specific comorbidity: coordination and continuity. Patient self-efficacy is an important mediating factor, so we will also assess patient activation.

- *Coordination of Care:* We will use the **Ambulatory Care Group (ACG) Coordination Risk Measure**,¹⁵ which considers the number of unique providers, the number of specialists, the percent of visits to the majority source of care and whether a generalist was seen to classify patients as having likely, possible, or unlikely risk of coordination problems. This measure was used in a previous R01,¹⁶ which assessed comorbid condition care in survivors of adult cancers. The overall risk classification will be our primary outcome for coordination of care, but we will also examine the four components of the measure individually.
- *Continuity of Care:* We will use the **Patient Perceived Continuity from Multiple Clinicians** to assess continuity of care.¹⁷⁻¹⁸ This 25-item measure includes three subscales related to the “responsible provider” and four subscales related to multiple clinicians and address continuity and coordination among the team. This generic instrument is applicable to a broad range of health conditions, including multimorbidity, and is appropriate for use in an ambulatory setting.

The Known Provider Continuity-Multiple Provider (KPC-MP) will be a secondary outcome.¹⁹ The KPC-MP is calculated as the total number of ambulatory care visits in Year X with all known providers seen in Year X-1, divided by the total number of ambulatory visits to all physicians seen in Year X.

- *Patient Activation:* We will use the 13-item **Patient Activation Measure® (PAM®)** to assess participant's knowledge, skill, and confidence for managing their health and health care.²⁰ The PAM categorizes scores into four levels of activation from low (1) to high (4). The measure has been validated in patients with a range of chronic conditions.

STUDY TEAM

Our team is exceptionally well qualified to conduct this study, having previously conducted research evaluating the quality of preventive and comorbid condition care in survivors of adult cancers, investigating the quality of care for populations with multiple morbidities, qualitative research with survivors and providers on cancer survivorship, disparities research, and health services research using the CCSS cohort.

- Principal Investigator:
 - Claire Snyder, PhD (cancer outcomes and health services research in cancer survivorship)
- Co-Principal Investigators:
 - Paul Nathan, MD, MSc (oncologist; survivorship clinical care and research)
 - Katherine Smith, PhD (survivorship research and qualitative methodology)
- Johns Hopkins Co-Investigators:
 - Cynthia Boyd, MD, MPH (geriatrician; multimorbidity research and clinical care)
 - Youngjee Choi, MD (general internist; survivorship clinical care and research)
 - Lorraine Dean, ScD (cancer and disparities research)
- CCSS Co-Investigators:
 - Greg Armstrong, MD (oncologist; survivorship clinical care and research; CCSS Principal Investigator)
 - Melissa Hudson, MD, (oncologist; survivorship clinical care and research)
 - Wendy Leisenring, ScD (statistician)
 - Aaron McDonald, PhD (CCSS Project Director)
- Advisors
 - Kevin Oeffinger, MD (family physician; survivorship clinical care and research)
 - Eric Chow, MD, MPH (oncologist; survivorship clinical care and research)

SUMMARY

The results of this study will (1) improve our understanding of the current patterns and quality of chronic condition care in

childhood cancer survivors, (2) identify the key socio-demographic factors influencing disparities in care, and (3) inform the design of optimal models of care for survivors.

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