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# Healthcare Utilization Among Young Adult Survivors of Childhood Cancer in Canada: The Role of Patient-Level Factors

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UNIVERSITY OF CALGARY

Healthcare Utilization Among Young Adult Survivors of Childhood Cancer in Canada: The Role  
of Patient-Level Factors

by

Rachelle Marie Drummond

A THESIS

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## Abstract

**Background:** Survivors of childhood cancer (SCC) face substantive risks for developing late effects (LE) from their cancer treatments due to disruptions in crucial periods of physical and social development while undergoing cancer treatment (Tonorezos et al., 2022; Baker & Syrjala, 2018). The inappropriate use of healthcare services, whether underutilization or overutilization, may jeopardize the effective risk-based prevention and management of LE among SCC. In addition, inappropriate healthcare utilization (HCU) may contribute to unsustainability and inefficiency in the healthcare system, creating challenges for supporting the growing population of survivors in Canada. Despite existing knowledge that young adult (YA) SCC are at a heightened high risk for the development of LE and life-long complications from their treatments, limited research exists that describes HCU among this population. The two aims of this study were to 1) identify and explore the associations of survivor's demographics (i.e., sex, gender, age, race, geographic location, SES), knowledge of cancer history and risks, and psychological factors (i.e., anxiety, depression) and perceived vulnerability with healthcare utilization; and 2) examine the relationships among demographic, clinical, and psychological patient-level factors with HCU among YA SCC.

**Methods:** Canadian YA SCC (n=123; 31% male; mean age=28.19yrs, mean time post treatment= 16.56yrs) diagnosed <18 years of age; and >5 years from diagnosis and/or >2 years from treatment completed the Self-Report Survey of Cancer Knowledge (Kunin-Batson et al., 2016), and the Self-Report Survey of Core Health Beliefs (Tercyak et al., 2004), to assess knowledge of cancer history and risks and perceived vulnerability, respectively. Survivors completed the Patient-Reported Outcomes Measurement Information System (PROMIS) SF v1.0 Anxiety 8a and the PROMIS SF v1.0 Depression 8a to assess levels of anxiety and depression respectively. To assess HCU, survivors reported the number of times that they saw a doctor within the last 2 years, in relation to their cancer diagnosis. A bivariate logistic regression model examined associations between YA SCC's demographics, knowledge of cancer history, and psychological factors with HCU.

**Results:** Survivors reported a wide range of HCU rates over the past 2 years (median= 3-4 visits). The number of reported visits ranged from 0-20+ visits. Despite 45.5% of reported health problems being attributed to mental health, psychiatrists and psychologists/counselors were the third and fourth least utilized types of providers, respectively. A logistic regression model was conducted to assess the associations between HCU and patient-related factors. The model was significant  $X^2(8) = 23.68$ ,  $p=0.003$ , and accounted for 29.3% of variance in healthcare utilization among YA SCC (Nagelkerke  $R^2 = 0.293$ ). Anxiety was the only independent variable that contributed significantly to the model ( $p<0.001$ ), and higher anxiety scores were associated with increased odds of HCU.

**Conclusion:** Anxiety levels were a significant predictor of HCU rates within this population, whereby higher anxiety scores were associated with higher HCU rates. Based on the findings of this study, behavioral interventions could be beneficial in reducing anxiety among YA SCC and promoting appropriate risk based HCU that support the long-term well-being of survivors, and the sustainability of the healthcare system.

## **Preface**

This thesis is an original, unpublished, and independent work conducted by the primary author, Rachelle M. Drummond. Dr. Fiona Schulte granted access to the dataset that was used, which was originally collected as part of a larger overarching study. The collection of data for this study was approved under the Health Research Ethics Board of Alberta- Cancer Committee (HREBA.CC-20-0248) on August 24, 2020.

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With sincere gratitude,  
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## List of Abbreviations

ASCC	Adult Survivor of Childhood Cancer
AYA	Adolescent and Young Adult
HCP	HealthCare Provider
HCU	HealthCare utilization
LE	Late Effects
PROMIS	Patient-Reported Outcomes Measurement Information System
PTSS	Post-Traumatic Stress Symptoms
QOL	Quality of Life
SCC	Survivors of Childhood Cancer
SES	Socioeconomic Status
YA	Young Adult



## **Chapter 1 General Introduction:**

Childhood Cancer and Young Adult Survivors of Childhood Cancer

## 1.1 | General Introduction

Substantial increases in the population of survivors of childhood cancer (SCC) in Canada can be attributed in part, to significant advances in treatment modalities and increased research efforts in the last few decades (Sharpless, 2021). As of 2021, there are an estimated 500,000 SCC residing in North America (Foster & Schulte, 2021). Such medical and technological advances have allowed a larger population of survivors to live longer than ever. In Canada, the 5-year survival rate for childhood cancer is approximately 84% (Public Health Agency of Canada, 2017), a 27% increase since the mid-1970's (American Cancer Society, 2024). The continuous growth of the population of survivors in North America necessitates a heightened focus on research aimed at developing novel approaches to improve access to risk-based healthcare services. These efforts are necessary to support the long-term needs of survivors while ensuring the sustainability of the Canadian healthcare system.

The completion of cancer treatment is often idealized as a finish line; however, it may signify the commencement of a challenging and lifelong journey for many survivors. Existing research suggests that this may be particularly true amongst individuals diagnosed with childhood cancer. As SCC often undergo invasive cancer treatments during critical periods of physical, cognitive, and emotional development, they are at a significant risk of developing late effects (LE) related to their cancer diagnosis and treatment (Brand et al., 2018; Baker & Syrjala, 2018). LE can be defined as health problems that occur months or years following the completion of cancer treatment and may be caused by the cancer itself or the cancer treatment modalities (i.e., surgery, chemotherapy, radiation therapy, or stem cell transplant) (National Cancer Institute [NCI], 2023). While LE may be broad and numerous, they can include chronic health conditions such as organ failure, hearing or vision loss, cognitive dysfunction, (Oeffinger

et al., 2006; Diller et al., 2009), secondary cancers (Bhakta et al., 2016), psychosocial difficulties (Hudson et al., 2003; Zebrack et al., 2004), fertility problems, and obesity (NCI, 2023; Hewitt et al., 2003).

## **1.2 | Childhood Cancer and Treatments**

Childhood cancer describes a group of cancers that are diagnosed in those from infancy to 18 years of age (Cleveland Clinic, 2023). Approximately 1,000 children are diagnosed with cancer each year in Canada (Statistics Canada, 2022). Childhood cancer incidence rates are notably higher among children at the younger end of the diagnostic age spectrum, with recent national statistics showing that children under the age of four are twice as likely to be diagnosed with cancer compared to those aged 5-14 years (Statistics Canada, 2022). In Canada, the three most common types of childhood cancer are: 1) leukemia (cancer of the blood and bone marrow) (31%); 2) central nervous system tumors (cancer of the brain and spine) (24%); and 3) lymphomas (cancer of the lymph nodes, bone marrow, and blood) (12%) (Statistics Canada, 2022). While the majority of cancers diagnosed in adults can be attributed to genetics, or lifestyle and environmental factors (World Health Organization [WHO], 2021) the origin of most childhood cancers are often unknown, making primary and secondary prevention challenging (Public Health Agency of Canada, 2017). Approximately 10% of childhood cancer diagnoses are thought to be attributed to genetic predispositions, but the remaining proportion remain unknown (WHO, 2021). Therefore, prioritizing the appropriate use and availability of health services is particularly crucial for tertiary prevention among children diagnosed with cancer.

Among the most common types of childhood cancer, the treatment of leukemia continues to have the most promising outcomes, with an average 94% survival rate (5 years post-

diagnosis) for those diagnosed between the ages of 0 to 14 (Canadian Cancer Society [CCS], 2023). Childhood cancer survival rates across all diagnosis types are expected to continue to improve as medical care and research focus increasingly on less-toxic, more targeted treatments, and advancements in clinical trials (Sharpless, 2021). Increases in survival rates will substantiate a larger population of SCC with unique and complex healthcare needs. To meet the needs of this population and optimize survivorship care, focused research efforts that aim to understand healthcare utilization (HCU) among SCC and the patient-level factors are required. HCU can be defined as the “the quantification or description of the use of services by persons for the purpose of preventing and curing health problems, promoting maintenance of health and well-being, or obtaining information about one’s health status and prognosis” (Carrasquillo, 2013). Globally, funding and research efforts remain focused on developing less toxic and invasive cancer treatments, with an emphasis on expanding immunotherapy clinical trials and advancing precision medicine to enhance long-term survivor wellbeing (Licciulli, 2023). While such efforts provide promise for improving survival rates and minimizing the impact of treatments on patients, complimentary efforts aimed at understanding the health service needs of survivors are required to support survivor’s long-term physical and psychosocial well-being.

### **1.3 | Late Effects in SCC**

While research suggests that almost all survivors of cancer will experience one or more LE from their cancer treatments, the severity and prevalence of such risks may be higher amongst those diagnosed with cancer as children. A review of global cancer burden among SCC (n=5,522) as part of the St. Jude Lifetime Cohort Study concluded that nearly 100% (95% CI 99.9%-99.9%) of SCC will develop at least one chronic health condition in their lifetime (Bhakta

et al., 2017) and upwards of 80% will experience a severe or life-threatening condition as a direct result of their cancer therapies and or diagnosis (Hudson et al., 2013). LE can have substantial impacts on a survivor's psychological and physical well-being, and may include increased risk of premature mortality, functional impairment, and impacts on health-related quality of life (QOL) (Suh et al., 2020; Hudson et al., 2013). Psychosocial difficulties from cancer treatments may include depression, anxiety, fear of recurrence, socialization difficulties, and post-traumatic stress (NCI, 2024; Hewitt et al., 2003). Additional studies have found that SCC are at risk for early onset of chronic disease, such as cognitive decline, and levels of physical frailty that are typically seen among older adults (Schuitema et al., 2021; Kruseova et al., 2023).

The risk of developing LE among SCC has been found to increase as time from cancer diagnosis elapses (NCI, 2024). This is due to the varying onsets among LEs and chronic health conditions (NCI, 2024). According to Mertens et al (2008), many LE and chronic health conditions may not emerge till 10 to 20 years post-treatment completion. A review of recent literature related to SCC's risk for subsequent malignant neoplasms, found that SCC over the age of 40 were found to have more than a twofold risk of developing secondary cancer in comparison to the general population (Turoctte et al., 2018). Similarly, incidence of cardiac events, including stroke, heart attack, and congestive heart failure, increased in survivors over the age of 35 (Armstrong et al., 2013). By 20 years of age, SCC are at an increased risk of developing severe, disabling, life-threatening, or fatal conditions from their cancer treatments compared to sibling controls (Armstrong et al., 2013). The delayed onset of LE highlights the importance of prioritizing tertiary prevention as an approach to promoting the long-term well-being of SCC.

## 1.4 | Young Adult Survivors of Childhood Cancer

The population of interest for this study is young adult (YA) SCC. Several personal and developmental characteristics establish this population as an important and unique participant group. The following section highlights some of these characteristics and their potential impacts on HCU.

### 1.4.1 *Disruptions in Development*

YA SCC are a unique population of study in that they undergo long periods of invasive and toxic treatments during critical periods of cognitive, physical, and social development (Brand et al., 2018). Disruptions during these stages of development place YA SCC at higher risk for LE and lifelong physical and social development challenges compared to age-matched controls and individuals diagnosed with adult-onset cancer (Tonorezos et al., 2022; Baker & Syrjala, 2018). Outcomes of cancer treatment, including prolonged hospitalizations and immunodeficiency, may contribute to disruptions in a patient's social development (Howard et al., 2014; Pahl et al., 2020). In comparison to sibling controls, SCC were found to demonstrate higher rates of social withdrawal, isolation, and loneliness, and to have difficulties getting along with peers (Pahl et al., 2020). Such disruptions could manifest into difficulties in developing social connections as survivors age (Howard et al., 2014; Pahl et al., 2020). These outcomes may decrease a patient's ability to socialize with peers or be in a communal setting (Freyer, 2010). Subsequent difficulties may arise in relation to navigating significant life changes and progressing through various stages of personal development, including the conceptualization of the pursuit of formal education, and the development of friendships and romantic relationships

(Freyer, 2010). Social-emotional development and maturity may also be affected by childhood cancer and its treatments through increased absenteeism from school and physical changes (i.e., hair loss, weight gain, scarring/physical disfigurement, etc.) (Mwanza, 2024). In a study that compared the psychosocial development of YA SCC with YA from the general population, it was found that survivors who had a central nervous system-based cancer were at risk for unfavorable psychosocial development across three identified psychosocial development domains (i.e., autonomy, psychosexual, and social development) (Maurice-Stam et al., 2022).

#### 1.4.2 *Medical Autonomy and Decision Making*

Canadian law and relevant provincial legislation stipulate that for most provinces and territories, anyone under the age of 18 is presumed to be legally incapable of consenting to medical care, unless otherwise specified under extenuating circumstances (Coughlin, 2018). In the case of pediatric populations, gaining informed consent typically involves gaining consent from a patient's legal guardian as a shared-decision maker (Coughlin, 2018). The authority to make medical decisions on behalf of a child requires a legal guardian/shared-decision maker to act in the best interest of a child under the age of 18 (Coughlin, 2018). Medical decision-making among adolescents and YA may be further complicated or impaired through psychosocial factors including peer pressure, impulsivity, and risk seeking behaviors that are not yet reconciled through physiologic maturation and life experiences (Coughlin, 2018).

When a patient is under the legal age of consent, information regarding the child's diagnosis, including treatment options and associated short-term and long-term risks are often communicated to the child's parents or legal guardians (Kadan-Lottick et al., 2002). Healthcare providers (HCP) are legally obligated to communicate treatment-related information to both the

child and the decision maker (Coughlin, 2018). However, the extent of information shared with a child may vary from that provided to the decision maker, depending on the child's level of understanding (Coughlin, 2018; Kadan-Lottick et al., 2002). Differences in the complexity or details of information shared between a HCP and a parent/decision-maker and that of a HCP and a patient may create knowledge barriers for survivors as they age and gain medical autonomy. In a cross-sectional survey of SCC (n= 635) from the Childhood Cancer Survivor Study (a multi-institutional cohort of adult SCC diagnosed between 1970 and 19886), only 15% (n=95) of participants reported having received a summary of the specific procedures and treatments they underwent, and the associated immediate and long-term risks (Kadan-Lottick et al., 2002). If YA SCC are unable to accurately recall or relay their medical history, their access to and appropriate use of medical care outside of their oncology team may be compromised. The availability of accurate and necessary medical information is arguably a prerequisite to receive appropriate and high-quality care (Bahrami et al., 2017).

Existing quantitative research has highlighted several factors that contribute to gaps in cancer history knowledge among YA SCC (Kadan-Lottick et al., 2002). A literature review that aimed to identify and discuss transitions from pediatric care to adult care among YA SCC highlighted gaps in knowledge about one's cancer history as a significant barrier to successful transitions in care (Freyer, 2010). As survivors age, they are faced with important changes and decisions related to their personal life, such as returning to school following cancer treatment, navigating peer relationships, and personal health risk management, all of which require accurate recall and understanding of their medical history and the risks they face later in life. YA survivors over 18 years of age and who are deemed capable decision-makers under regional law are granted autonomous decision-making capabilities for all health-related decisions, including



cancer care. It is hypothesized that the shift from relying on a legal guardian/shared-decision maker for decisions related to an individual's healthcare, to making one's own decisions, may contribute to stress and confusion for YA SCC. Although survivors are responsible for making decisions about attending cancer-related appointments, system-level factors such as accessibility of care, availability of resources, and geographical location may hinder their ability to attend, even if they intend to do so. Nevertheless, the identification of patient-level factors that may influence a YA SCC's decision or ability to utilize healthcare services can serve as a valuable point of intervention in future work aimed at promoting appropriate risk-based healthcare utilization (HCU) among YA SCC.

#### 1.4.3 *Transitions from Pediatric to Adult-Centered Care*

The transition from being a pediatric patient to a YA SCC can be a significant shift for many individuals as they go from being dependent on parental decision making, to independent active decision makers in their own regard (Bahrami et al., 2017). It can be hypothesized that in instances where a survivor has newly granted medical autonomy and is concurrently tasked with transitioning from pediatric to adult-centered care, this may contribute to differences in survivorship experiences and healthcare use among SCC. A recent systematic review of studies on transitions from pediatric to adult follow-up care among SCC found that the most reported barrier across all included studies (n=26) was SCC's self-management skills (Otth et al., 2021). Several different variables were considered within the self-management skills of survivors, such as their knowledge of their cancer history and risks, their comfort with and knowledge of the adult care system, and their psychological well-being following treatment (Otth et al., 2021). A proactive approach to educating survivors about the transition from pediatric to adult care could

enhance their understanding of survivorship services and lead to improved utilization of these services (Otth et al., 2021). In contrast, a qualitative study from Australia examined the attitudes and experiences of SCC relates to transitioning from pediatric to adult care and found that many survivors expressed their medical independence and self-management skills as an enabler to care (Nandakumar et al., 2018). Survivors noted the transition as being a way to be less dependent on HCP and parents to navigate their medical care, empowering them to go on their own, and reducing the attachment to and reliance on their childhood care team (Nandakumar et al., 2018).

Common barriers noted above related to knowledge deficits, reliance on parents or existing teams of HCPs, age, and cognitive deficits may contribute to improper utilization of survivorship healthcare services among YA SCC. Further research is needed to understand the challenges that participants have faced throughout their cancer journey. Such information can be beneficial in informing current patients, HCPs, and health systems experts of potential challenges, with the goal of developing health systems change and interventions aimed at preventing these challenges for future generations of survivors.

## **Chapter 2: Literature Review:**

Healthcare Utilization Among Young Adult Survivors of Childhood Cancer

## 2.0 | Healthcare Utilization

Understanding HCU among SCC is helpful for conceptualizing the optimal delivery of survivorship care services, however, the under or overutilization of services is not well substantiated in literature. The quantity and type of healthcare services an individual uses, and their access to care, can vary due to several factors at both the individual and system levels (Meyers, 2008). Individual-level factors include a person's health status, their service needs, their gender, their SES, and their individual decision-making style (Meyers, 2008). Alternatively, system-level factors may include the availability of resources within a specific healthcare system including staffing, funding, and capacity considerations (Meyers, 2008).

It is important to consider differences in HCU through a comparative lens that recognizes differences in healthcare systems across countries. Within the publicly funded Canadian healthcare system (i.e., Medicare), Canadians are provided with insurance that covers all medically necessary services provided by hospitals, physicians, and dentists (when the service must be provided in the hospital) (Government of Canada, 2023). In most instances, treatments, procedures, and prescriptions for children diagnosed with cancer in Canada are covered through Medicare (Canadian Cancer Society, 2024<sup>B</sup>). The United States has a mixed system, where there is not universal coverage, but rather the combination of government-funded Medicare (for those 65 years and older), Medicaid (for low-income individuals, pregnant women, and children) and privately funded insurance (ISPOR, 2024). Despite the opportunity for coverage of cancer-related care through state-funded Medicaid, this coverage only applies to those under the age of 19 (unless otherwise pregnant or within the low-income threshold) (United States Government,

2024), leaving many YA SCC without guaranteed insurance, and at risk for access barriers to crucial health services.

Most research related to HCU among cancer survivors either pairs YA SCC with adolescents and children or is centered around individuals diagnosed as adults. While these studies do not typically classify usage as under- or overutilized, they do report high HCU rates among survivors. This study seeks to close this gap by assessing the association between survivor demographics (i.e., sex, gender, age, race, geographic location, SES), knowledge of cancer history and risks, and psychological factors (i.e., anxiety, depression, and perceived vulnerability) with HCU related to one's cancer diagnosis over a 2-year period. Particularly, we are interested in knowing which survivor demographics or patient-level factors increase or decrease HCU rates and which healthcare providers are utilized the most by YA SCC.

## **2.1 | Risk-Based Healthcare Utilization**

For cancer survivors, the development of a survivorship care plan is typically guided by a risk-based care approach (McCabe et al., 2013). Risk-based care can be described as the development of a survivorship care plan that involves screening, prevention, and surveillance strategies that consider a patient's individual health risks, as predisposed by cancer history, genetics, family history, lifestyle factors, and comorbidities (McCabe et al., 2013). A risk-based approach to HCU may be difficult for providers or survivors to achieve if the diagnostic history and associated risks of an individual are unknown or are not understood (McCabe et al., 2013). Gaps in knowledge pertaining to one's cancer history and their associated risks may result in the inappropriate use of healthcare services. The term "inappropriate healthcare use" encompasses both the underutilization and overutilization of healthcare services (Squires et al., 2019). Both

instances may not only pose risks to the well-being of healthcare system users, but also to the overall sustainability and efficiency of the healthcare system (Squires et al., 2019). For health system users, such as YA SCC, inappropriate healthcare use can lead to negative patient experiences and poor outcomes (Squires et al., 2019). These adverse outcomes may stem from the underutilization of effective clinical practices or services (i.e., recommended risk-based screening), or from the overuse of harmful/ineffective services (i.e., unnecessary testing) (Squires et al., 2019). For the healthcare system itself, instability in the system may be revealed when high rates of HCU push a system past its capacity.

An example of instability in the healthcare system recently occurred in Alberta, where the onset of some chemotherapy treatments for childhood cancer patients were delayed due to health system capacity challenges (Moulitharan, 2023). According to the Alberta Medical Association, long wait-times for emergency care, staff shortages, and capacity challenges are part of the reason that essential medical care for children is being impacted (Moulitharan, 2023). Such insights highlight the interconnectedness of the healthcare system and the associated trickle-down effect of complexities in one health area onto another. Despite our understanding of the unique experiences of YA SCC and the extensive long-term risks for LE that they face, limited research is available that examines HCU among YA SCC as a standalone population of interest. Overutilization of healthcare services could cause unnecessary harm to survivors or contribute to strain on the health system. Conversely, underuse of healthcare services could negatively impact the long-term QOL of survivors if they do not receive the appropriate risk-based care that is required to prevent or reduce the risk of LE and co-morbidities resulting from childhood cancer treatment.

## 2.2 | Guidelines for Healthcare Utilization

Survivorship guidelines are commonly used in Canada as a tool to help guide survivorship care and to improve knowledge of risks among SCC. The Children’s Oncology Group “Long-term follow-up guidelines for survivors of childhood, adolescent, and young-adult cancer” were developed for clinicians as a means of standardizing and improving survivorship care practices and providing recommendations for the management and prevention of LE (Children’s Oncology Group [COG], 2023). The COG states that while designed for clinical practice, their guidelines can be a valuable tool for survivors if they are reviewed by a survivor with the assistance of a healthcare professional that is knowledgeable about long-term follow-up care (COG, 2023). In addition, “healthlinks” have been integrated into this document that allows survivors to access patient education materials. Documented shortages within the Canadian oncology workforce (Chaput, 2023) may present challenges for survivors in accessing healthcare providers adequately trained in follow-up care who can devote sufficient time to thoroughly explain and review the COG guidelines in a manner comprehensible to survivors. When considering appropriate HCU among YA SCC, it is important to consider what information on available healthcare services is accessible to survivors, and the impacts that it could have on their HCU rates.

Efforts to improve accessibility to survivorship services and to empower survivors in their care in a more modern way, have been seen through the creation of digital navigation and health tracking tools for survivors. The “passport for care” is an American-based free online resource for SCC that was designed in collaboration and alignment with the COG (Passport for Care [PFC], 2024). Survivors are provided with a unique code by their healthcare provider that allows them to create an account on the passport for care website, where they can access their

treatment summary and a tailored survivorship care plan/recommendation (PFC, 2024). All information available to a survivor can be printed and/or shared with a healthcare provider, at the survivor's discretion. The use of tools such as the passport for care varies greatly globally. In Canada, some provinces have begun to adapt the passport for care for use in the Canadian healthcare system. Eastern provinces under the jurisdiction of Eastern Health Cancer Care have a "patient passport" (Newfoundland) or a "oncology passport" (Quebec) that is available to cancer patients and survivors, but these passports are not available in a digital format, such as a website or application. Inconsistencies in the accessibility and availability of information for SCC may contribute to improper healthcare service use among this population. Continuous growth in the global and North American populations of SCC substantiates the crucial need to identify and conceptualize the unique healthcare needs of this population. This information will offer valuable insights for developing standardized survivorship care and contribute to a more effective and sustainable healthcare system that can support the growing survivor population, promoting their long-term well-being through appropriate, accessible, and efficient healthcare service use.

### **2.3 | A Conceptual Frameworks for HealthCare Utilization**

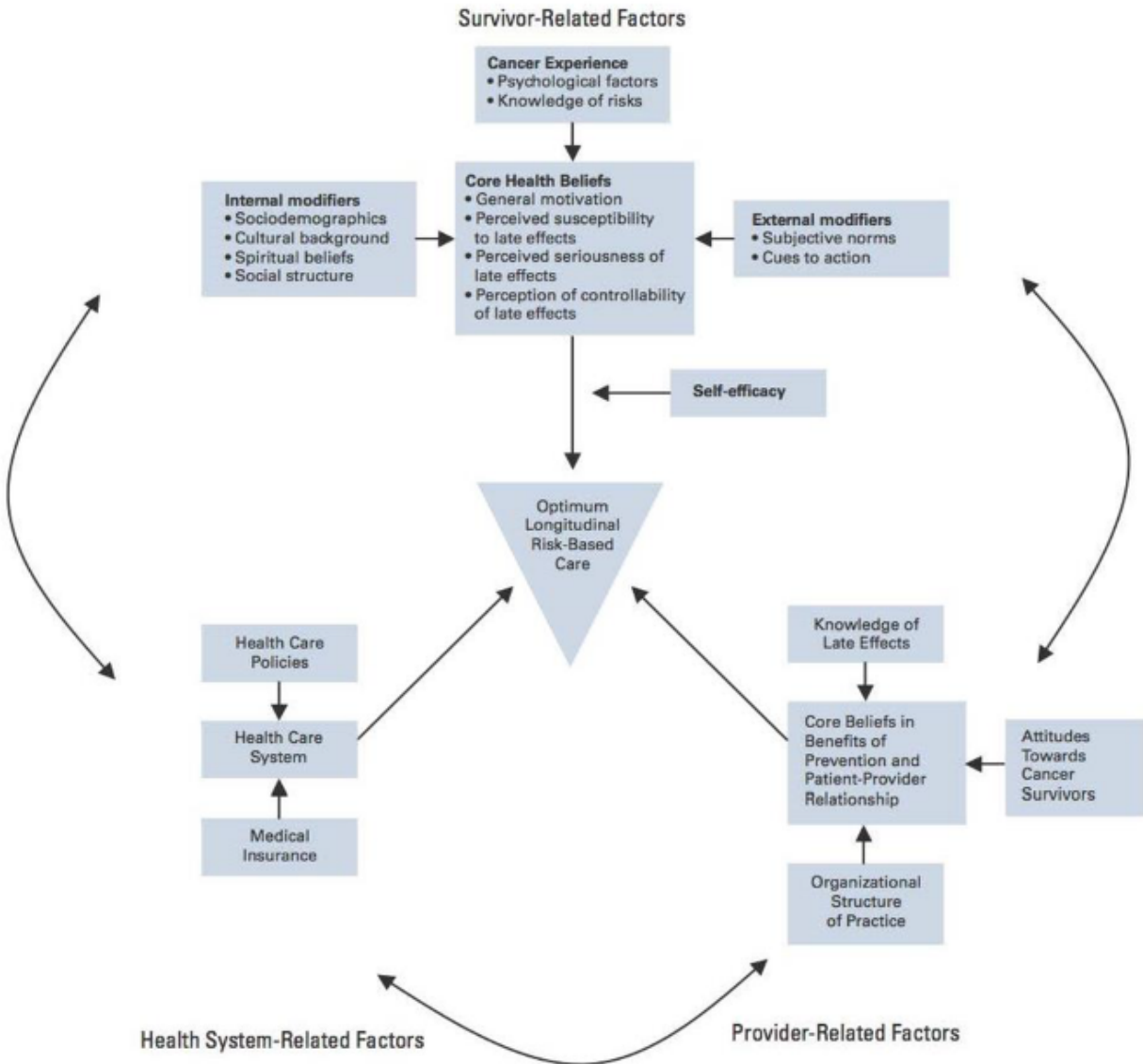
In recognition of the importance of appropriate HCU for the long-term well-being of SCC and the overall sustainability of the healthcare system, it is crucial to understand the complex interplay between the unique healthcare needs of YA SCC, patient-level factors, and HCU. A conceptual model will provide insight into patient-level factors that are key to promoting appropriate HCU that considers the unique complexities and experiences among our study population.



Conceptually, HCU has been described using several theoretical models, however, to our knowledge, there are no conceptual frameworks that explore HCU in the context of childhood cancer. The following section highlights a theoretical model by Oeffinger (2003) that identifies survivor-related barriers and enablers to achieving optimum longitudinal risk-based care. Through this model, we gain insight into the role of survivor's cancer experiences, health beliefs, and personal factors in relation to HCU.

**Figure 1**

*Theoretical model of potential barriers and enablers to the longitudinal cancer-related healthcare of adult survivors of childhood cancer (Oeffinger, 2003)*



### 2.3.1 *Barriers and enablers to longitudinal cancer-related healthcare*

Oeffinger (2003) developed the theoretical model of potential barriers and enablers to the longitudinal cancer-related healthcare of adult SCC by integrating the Health Belief Model (Rosenstock, 1974), the Health Locus of Control Model (Rotter, 1954), and the Behavioral Model of Utilization (Andersen, 1995; Philips et al., 1998). To examine the relationships between 1) survivors; 2) the healthcare system; and 3) healthcare providers, Oeffinger's theoretical model considers barriers and enablers to care across each of these 3 domains, and their association with optimum longitudinal risk-based care. As described by Oeffinger (2003), survivor-related barriers and enablers can be grouped into five sub-domains: 1) the cancer experience; 2) core health beliefs; 3) internal modifiers; 4) external modifiers; and 5) health locus of control. Healthcare provider-related barriers and enablers are further categorized into four sub-domains: 1) the core health beliefs of the HCP regarding the value of prevention; 2) the provider's knowledge of LE and long-term risks faced by survivors; 3) attitudes towards cancer survivors; and 4) the organizational structure of the practice. Lastly, healthcare system-related barriers and enablers are further categorized into three sub-domains: 1) medical insurance; 2) the healthcare system; and 3) healthcare policies regarding survivors. The theoretical model of potential barriers and enablers has been validated and widely used to examine barriers and enablers to care across several cancer diagnosis and study types, including the development of clinical care guidelines (Tonorezos & Henderson, 2014).

The cylindrical structure of this framework highlights the crucial interplay between survivor-related factors, health system-related factors, and provider-related factors in achieving optimal longitudinal risk-based care. According to Oeffinger (2003), this theoretical model can be used as a framework to develop and test interventions aimed at improving care for SCC. This

study will focus on survivor-related factors and enablers within this model, specifically examining the core health beliefs outlined by Oeffinger.

An individual's core health beliefs include: 1) general motivation towards health; 2) perceived susceptibility to disease (or late-effects); and 3) perceived seriousness of disease or late-effects (Oeffinger, 2003). As defined in the health belief model (Rosenstock, 1974), perceived susceptibility refers to a person's subjective perception of the risk of acquiring an illness or disease (in this case, secondary cancer, or late-effects/chronic disease), and perceived seriousness refers to a person's feelings on the seriousness of contracting an illness or disease, and often is based on a person's considerations of the medical and social consequences of contracting an illness or disease. This domain will help guide our understanding of survivors perceived vulnerability to LE and long-term risks.

Considering the significant long-term morbidities of LE and chronic health conditions in the YA SCC population (Bhakta et al., 2016) and the role of risk-appropriate HCU in mitigating the risk of morbidities, it is imperative to understand the impact of patient-level factors on HCU. The application of Oeffinger's theoretical model will allow us to conceptualize and understand the complex interaction between survivors' experiences and their healthcare needs. This information may help inform future interventions and policy/health system changes aimed at improving healthcare service access and quality among YA SCC.

#### **2.4 | Patient-level Factors Related to Healthcare Utilization**

Unsurprisingly, more frequent utilization of healthcare services is consistently seen among SCC compared to non-cancer survivors across all areas of health service utilization, including mental health services, in-patient and out-patient services, and emergency services (Miser et al., 2023; Rebholz et al., 2011). Findings from the British Childhood Cancer Survivor

Study, a population-based cohort study of individuals diagnosed with childhood cancer (n=17,891) and surviving  $\geq 5$  years, suggested that while contact with a general practitioner was similar among survivors and controls over a 2-week period, SCC tended to use in-patient, out-patient, and specialized care more than a control group of the public (when controlling for regular follow-up care attendance) (Rebholz et al., 2011). Given the short-term and long-term complexities of a cancer diagnosis, it is unsurprising that this population has been found to have higher rates of HCU in comparison to the public. However, it is important to quantify and understand HCU rates among a population of YA SCC to better understand their healthcare needs. This study is interested in identifying and understanding patient-related factors unique to YA SCC that may impact their levels of HCU in comparison to others within the same population.

#### 2.4.1 *Sex and Gender*

As outlined in Oeffinger's (2003) theoretical model, an individual's assigned sex and their chosen gender may play an important role in their health behaviors, and subsequent outcomes. Research on self-reported HCU and sex among SCC has largely concluded that individuals of female sex tend to have higher HCU rates in comparison to self-reported HCU among individuals of male sex (Oeffinger et al., 2004; Castellino et al., 2005). In a retrospective cohort study of adult SCC from the Childhood Cancer Survivor Study (n=9,434), baseline self-report questionnaires were utilized to ascertain participant's cancer-related medical visits (as well as gather participant demographic and clinical information) (Oeffinger et al., 2004). Univariate regression analysis identified an association between male sex (OR= 1.65; 95% CI, 1.44-1.88) and reporting a general physician examination, a cancer-related visit, or a cancer

center visit (Oeffinger et al., 2004). To our knowledge, no studies exist that specifically look at the relationship between gender and HCU within a population of YA SCC. The absence of gender based HCU research is a matter of concern and warrants further emphasis for several reasons. First, sex and gender are often conflated in research, where they are used interchangeably. The validity and generalizability of findings from studies that do not distinctly differentiate between sex and gender is limited. Second, the absence of gender-based research may contribute to challenges for future studies or interventions that primarily target gender differences if it is unclear whether existing studies conflated sex with gender.

Sex and gender differences may arise due to differences in biological, behavioral, and social factors. Biological factors related to one's sex at birth can increase the risk for certain cancer diagnoses and the development of certain LE (Oertelt-Prigione et al., 2021). For example, in directly comparing sex differences between male and female long-term cancer survivors in symptoms and functioning with an age-matched reference population, individuals of male sex were found to experience significant long-term impacts, such as significant net loss in role and social functioning ( $-9.9$  [95% CI =  $-11.2$ ;  $-8.6$ ] and  $-7.7$  [95% CI =  $-9.6$ ;  $-7.6$ ] respectively). While sex largely influences cancer biology and outcomes (Lopes-Ramos et al., 2020; WHO, 2021<sup>B</sup>), gender norm and roles may impact one's experiences with healthcare, the way they interact with the healthcare system, their access to care, and their perceptions of their cancer journeys (WHO, 2021<sup>B</sup>).

Despite the complexities in the interactions between sex, gender, and health outcomes, there are significant limitations in the available research that accurately distinguishes between sex and gender differences, as the two terms are often used interchangeably. Without a foundational understanding of gender based HCU research, there is limited research to

substantiate findings. This study will analyze both sex and gender in relation to HCU among YA SCC. This is imperative to not only address existing research gaps, but to improve the generalizability and validity of findings when being applied across different sex and gender populations of YA SCC.

#### 2.4.2 Age

Most of the existing literature on age and YA SCC concludes that as survivors age, they are less likely to report using healthcare services and are more likely to have lower HCU rates (Oeffinger et al., 2004; Castellino et al., 2005). A retrospective cohort study of YA SCC from the Childhood Cancer Survivor Study (n=9,434) identified that the likelihood of self-reporting a cancer-related visit or general physician examination decreased significantly ( $p < 0.001$ ) as a survivor aged (Oeffinger et al., 2004). Similarly, a quantitative study that aimed to determine the influence of race and ethnicity on the outcomes in the Childhood Cancer Survivor Study, found that regardless of race or ethnicity, childhood cancer survivors as a population may face impaired healthcare access as they age, due in part to socioeconomic status (Castellino et al., 2005). Such findings may have critical clinical implications given existing research suggests that the incidence and risk for LE and chronic conditions increases as survivors age (Oeffinger et al., 2004). An increase in the incidence of LE, coupled with a decrease in risk-based care as survivors age, underscores the need to reconsider and potentially revise current survivorship care standards. This study will contribute to existing literature on age and HCU, which may help inform care standards for YA SCC.

### 2.4.3 Race

Few studies were identified that analyzed associations between race and HCU among YA SCC. A report from the Childhood Cancer Survivor Study analyzed HCU, long-term outcomes, and health-related behaviors among Black and Hispanic adult SCC (Castellino et al., 2005). Logistic regression analysis revealed that, compared to White survivors, Hispanic survivors (male and female) were more likely to report a cancer center visit (females: OR = 1.5; 95% CI, 1.1-2.0; males: OR = 1.7; 95% CI, 1.2-2.3) and had more equitable access to cancer related care than Black or White survivors (Castellino et al., 2005). The authors hypothesized that participation in childhood cancer protocols and the resultant familiarity with the medical system may have mitigated some of the traditional barriers to healthcare faced by racial minority groups (Castellino et al., 2005). When adjusting for SES, self-reported utilization patterns over a 2-year period were similar across racial groups (Blacks, Hispanics, and Whites) (Castellino et al., 2005), suggesting that system-level factors, such as income, may be a significant barrier to accessing healthcare services among racial minority populations.

When considering the impacts of sex differences on self-reported HCU rates across racial groups, Black males were less likely to report general medical contact (OR= 0.60; 95% CI, 0.5-0.9) in comparison to adult SCC of White or Hispanic race when not adjusting for SES (Castellino et al., 2005). Irrespective of SES, Black female and Hispanic male survivors had less general contact with the medical system yet had equitable use of cancer-related services in comparison to White survivors (Castellino et al., 2005). Further, in comparison to White survivors, both male and female Hispanic survivors were more likely to report a visit to a cancer center (Castellino et al., 2005). As described previously (see section. 2.2.1), authors hypothesized that childhood cancer and the associated need for access to healthcare services may



remove some of the traditional barriers to healthcare experienced by racial minority groups in comparison to the general population.

The absence of research examining the association between race and HCU among YA SCC complicates the development of effective population-specific interventions aimed at improving equity in HCU for YA SCC. Thus, it is essential to gather insights into the association between HCU and race within our population and in the Canadian context to contribute to identified research gaps.

#### *2.4.4 Psychological Factors*

As substantiated by the theoretical model of barriers and enablers, psychological factors may have an interconnected relationship with HCU and optimal risk-based care (Oeffinger, 2003). These factors are unique in that they are often modifiable through interventions such as therapy, psychiatric services, and other psychological supports (Zhang et al., 2022; Coughtrey et al., 2018) The modifiable nature of these factors provides potential for minimizing inequitable or inappropriate risk-based HCU through evidence-informed interventions.

In a quantitative report from the Childhood Cancer Survivor Study, Crochet et al., (2019) utilized modified Poisson regression and multinomial logistic regression models to analyze the associations between post-traumatic stress symptoms (PTSS) and health outcomes among adult survivors of childhood cancer (ASCC) (n=6,844). After adjusting for demographic and disease-related variables, survivors with PTSS (as determined by the Posttraumatic Stress Scale) were found to be more likely than survivors without PTSS to have reported no primary care visits in the past 2 years (39% of survivors with PTSS in comparison to 33.7% without) (Crochet et al., 2019). However, 28.2% of survivors (n=1,930) with PTSS reported attending more than seven

medical visits in the last 2 years, in comparison to 20.1% (n=1,376) without PTSS (Crochet et al., 2019). Survivors with hyperarousal and intrusive symptoms of PTSS utilized higher rates of general healthcare (OR=1.04, 95% CI= 1.00-1.08) (Crochet et al., 2019). In analyzing the number of cancer-related visits among those with and without PTSS, survivors with PTSS were more likely to have had a cancer-related visit in the past 2 years (42.2%) in comparison to survivors without PTSS (29.1%) (RR = 1.31, 95% CI = 1.18–1.46,  $p < 0.0001$ ) (Crochet et al., 2019). Survivors in the St. Jude Lifetime Cohort Study, a retrospective cohort of SCC treated at St. Jude hospital (n= 3,211) completed self-report questionnaires to assess their cancer-related worry, health behaviors, and psychological and somatic symptoms (i.e., anxiety and depression) (McDonnell et al., 2021). Results of multivariable regression models indicated that cancer-related worry (i.e., concerns about relapse and LE) was associated with higher rates of HCU when chronic conditions and psychosocial variables were controlled for (McDonnell et al., 2021).

The abovementioned studies highlight several interesting findings that are relevant to our population of interest. First, as highlighted by Crochet et al., (2019) while survivors with PTSS are more likely to engage in and use cancer-centered care than those without PTSS, their use of primary care exhibited extremes, where survivors were either highly engaged (7+ visits in a 2-year period) or completely disengaged (0 visits in a 2-year). Both McDonnell et al. (2021) and Crochet et al. (2019) highlighted the interplay between psychological LE of cancer (including cancer-related worry, PTSS) and HCU rates. Differences in primary care versus cancer-related care utilization within this population could be attributed to several complex and patient-level factors such as trust between patients and providers, knowledge of available services, perception towards disease and risk, and accessibility challenges (Crochet et al., 2019). Further, given that

these studies were conducted within the US, differences in utilization between primary care and specialized care may have been related to cost differences across primary care and specialized care (see section 2.0). Varying levels of cancer-related worry, anxiety, or PTSS may either act as a driving force or enabler for survivors to utilize higher levels of care as a means of calming their health-related anxiety, or as a barrier, whereby they may be fearful or anxious of test results or HCPs in general.

#### 2.4.5 *Knowledge of Cancer History and Associated Risk*

A patient's knowledge of their cancer history and the associated risks may be associated with their levels of HCU (Gianinazzi et al., 2022). Disruptions caused by cancer treatments during periods of development (Brand et al., 2018; Baker & Syrjala, 2018) may place YA SCC at greater risk for developing LE throughout their lifetime. However, survivors may be unaware of these risks and how to minimize them without knowledge of their cancer history. Insufficient knowledge related to one's cancer history may arise for several reasons. As mentioned previously, the role of parental decision-making for patients under the age of 18 may create gaps in knowledge for patients who received little to no information regarding their diagnosis or long-term risks (Coughlin, 2018). Additional factors related to gaps in knowledge may be psychological in nature (Kadan-Lottick et al., 2002; Murphy et al., 2021). Several examples of existing research note the connection between factors such as age at diagnosis and psychological trauma with recall difficulties. A cross-sectional survey of CCS (n=635) (Kadan-Lottick et al., 2002) (2002) utilized telephone surveys to assess survivors' knowledge of their cancer history. This self-reported information was then compared to survivor's medical history to determine the accuracy of their knowledge. While 74% of survivors were able to provide an accurate *general*

summary, no participants were able to provide an accurate *detailed* summary (Kadan-Lottick et al., 2002). Similarly, in a randomized study of CCS (n=126), participants were randomized to receive or not receive a survivor healthcare passport at their first survivorship care appointment (Murphy et al., 2021). After assessing survivor's knowledge of cancer history at three timepoints using a questionnaire, it was found that those who immediately received their passports demonstrated improved and sustained knowledge over a four-month period in comparison to those who did not receive the passport immediately (Murphy et al., 2021). Further, in instances where survivors were provided with information of their cancer history and risks, they may have difficulties in recalling such information due to various factors such as trauma-induced recall difficulties or some information being shielded from them by their parent/caregiver (KadanLottick et al., 2002), These studies emphasize the importance of providing survivors with detailed summaries of their cancer history and risks early in survivorship. It is also crucial to ensure that this information remains readily available for reference to minimize recall difficulties.

Existing research relating to knowledge of cancer history and HCU is predominantly centered around the underutilization of cancer-related services among survivors. While this literature is important for guiding follow-up care utilization and capacity, further research is required to understand how survivor's cancer knowledge translates to non-cancer-related service use and how knowledge of future risks may contribute to the under or over utilization of healthcare services. A report from the Childhood Cancer Survivor Study on outpatient medical care among YA SCC concluded that one of the key obstacles contributing to underutilization of cancer-related care services was gaps in survivors' knowledge of their cancer diagnosis and long-term risks (Oeffinger et al., 2004). Authors state that while most survivors have general

knowledge regarding their cancer diagnosis and the type of treatment they received, very few had an actual summary of their treatment history or were aware of the risks and LE that they face from their treatments (Oeffinger et al., 2004).

The limited findings related to HCU and knowledge of cancer history and risks among YA SCC highlight the need for additional analysis and research in this area. This study will analyze survivor's self-reported knowledge of cancer history and risks as a contributing factor to HCU, across both general and cancer-related care. Such information may contribute to future interventions aimed at improving knowledge translation and sharing among patients, survivors, and HCPs. Through improved knowledge translation, it is hypothesized that survivor's HCU may become more risk-appropriate for their individual needs.

#### *2.4.6 Perceived Vulnerability*

As described in Oeffinger's theoretical model (2003), perceived vulnerability or perceived susceptibility to disease is highly variable based on an individual's perception of their risk. Such internal processing may be a critical factor in a survivor's health behavior choices. As such, it is essential to understand the role of perceived vulnerability in relation to HCU. Such information may help inform future educational interventions aimed at helping survivors develop an appropriate risk-based perception of their susceptibility to disease.

No studies were identified that directly analyzed the association between perceived vulnerability and HCU among YA SCC. A retrospective cohort study of adult SCC (n=9,343) by Oeffinger et al., (2004) utilized four self-reported outcome measures to measure outpatient medical care in a 2-year period, including 1) general contact with the healthcare system; 2) general physician examinations; 3) cancer-related medical visits; 4) and medical visits at a cancer-center. A lack of concern for future health was identified as a factor associated with not

reporting a general physician examination, cancer-related visit, or a cancer center visit (OR= 1.57; 95% CI, 1.36-1.82) (Oeffinger et al., 2004). Based on such results, it could be hypothesized that those who have low levels of concern for their future health may not perceive that they are highly vulnerable to future disease or illness.

Given our understanding of the role of perception and individual decision-making on health behaviors (Oeffinger, 2003), it is necessary to minimize this identified research gap to help promote evidence-informed decision making for survivors in relation to their healthcare. As such, this study will analyze the relationship between perceived vulnerability and HCU among YA SCC.

## **2.5 | Gaps in Literature**

While there is a growing body of research relating to HCU among SCC, we identified two substantial gaps in existing research that we aim to address through this study. First, HCU research for YA SCC predominantly exists as a sub-group alongside adolescents in the AYA population. As mentioned previously (see section. 1.4.1), YA SCC are a unique population of study. They often experience prolonged, invasive, and toxic cancer treatments during crucial periods of physical, social, and cognitive development (Brand et al., 2018; Baker & Syrjala, 2018), coinciding with the critical transition from childhood to adulthood, a time that demands increased autonomy and involvement in medical decision-making (Bahrami et al., 2017). Such interruptions have been found to place them at greater risk for morbidities of LE and chronic conditions as they age (Oeffinger et al., 2004). Generalizing HCU research findings from an AYA or adult population of SCC to a YA population of SCC would conjure issues of validity given our understanding that these populations are distinct.

Secondly, existing studies on HCU among YA SCC have been conducted predominantly in the US, where public healthcare services/facilities are limited. Significant differences in access to healthcare services exist between the U.S private healthcare system and Canada's public healthcare system. These disparities limit the applicability of findings from American-based HCU research in a public healthcare context with fewer cost-related barriers. To further substantiate the inability to directly apply findings from a private healthcare system to one that is public, most existing literature on HCU among SCC analyzes American healthcare insurance as a primary predictive factor for service use. Given that YA SCC in Canada would have medical coverage under the provincial healthcare system, this information is not relevant to our population of interest and cannot be generalized to the Canadian healthcare system.

This study will aim to contribute to existing gaps in YA SCC research related to HCU by applying a cross-sectional quantitative approach to analyze underrepresented patient-level factors in the literature, including, knowledge of cancer history and risks, perceived vulnerability, gender, and race. Findings will offer new insights into HCU among YA SCC in a public healthcare setting and highlight YA SCC as a standalone population of interest, which is essential for helping to inform population-appropriate healthcare services in the Canadian context.

## **2.6 | Research Aims and Hypotheses**

**Aim 1:** Identify and explore the associations of survivor's demographics (i.e., sex, gender, age, race, geographic location, SES), knowledge of cancer history and risks, and psychological factors (i.e., anxiety, depression, and perceived vulnerability) with healthcare utilization among young adult survivors of childhood cancer.

**Hypothesis 1:** For aim 1, we hypothesize that there will be a range of HCU reported among our sample and differences seen between groups reporting high vs. low HCU rates, it is hypothesized that significant associations will exist between anxiety and depression.

**Aim 2:** Examine the relationships among demographic, clinical and psychological patient-level factors with healthcare utilization among young adult survivors of childhood cancer.

**Hypothesis 2:** For aim 2, it is hypothesized that lower healthcare utilization will be associated with male sex and gender, rural/remote living, lower SES, older attained age, higher anxiety scores, and lower perceived vulnerability.



## **Chapter 3: Methods**

### **3.1 | Participants**

Survivors were eligible to participate in this study if, at the time of the study they were: (1) between the ages of 18 to 39 years; (2) diagnosed with cancer under the age of 18 years; (3) at least 5 years from initial diagnosis and/or two or more years post-treatment completion; and (4) currently living in Canada. These inclusion criteria are consistent with the COG definition of a survivor (ASCO, 2022). Individuals who did not satisfy the abovementioned inclusion criteria or were not able to read English were excluded from the study. No further restrictions were placed on eligibility to ensure the representativeness of the sample and ecological validity of the findings.

### **3.2 | Recruitment**

Several methods of recruitment were employed throughout this study to gather a diverse and representative sample of YA SCC. Ethics approval for this study was granted by the Health Research Ethics Board of Alberta Cancer Committee (HREBA.CC-20-0248). Recruitment was conducted between September 2021 and December 2023.

#### *3.2.1 Primary Recruitment Methods: Long-Term Follow-up Clinics*

Primary recruitment was conducted through Canadian LTFU care clinics. During regularly scheduled visits, eligible survivors (as identified by the research team) were notified by their local LTFU clinic care team of the opportunity to participate. At this time, LTFU clinic care team members provided interested survivors with a study information sheet and consent-to-contact form. Individuals who completed the consent-to-contact form were contacted by a member of the research team to confirm interest and eligibility. Those confirmed to be eligible

by a research team member were emailed a unique link to complete the online study questionnaires by a research team member.

### *3.2.2 Secondary Recruitment Method: Patient Partner Recruitment*

Patient partners from the Psychosocial Care4Kids Lab (n=4) assisted in the recruitment of additional participants across Western, Central and Eastern Canada through both personal relations and local LTFU clinic outreach. Peer-to-peer recruitment is an important principle in support of the inclusive mechanisms and processes outlined by the CIHR SPOR Strategy for Patient-Oriented research (2014). We inferred that the use of patient partners in recruitment helped to foster trust among potential participants through mutual understanding and shared experiences.

### *3.2.3 Tertiary Recruitment Method: Social Media Strategy*

Ongoing recruitment utilized Psychosocial Care4Kids lab social media accounts (i.e., X/Twitter, Facebook, and Instagram). Social media recruitment was intended to help facilitate recruitment of survivors from across Canada who were not attending LTFU care clinics (or who were not scheduled for an appointment during the study period).

## **3.3 | Study Design and procedures**

This thesis project is part of a larger multi-phased mixed-method study that aims to develop an mHealth intervention to improve follow-up care experiences for SCC. To support the aims of this study, only quantitative data pertaining to variables of interest were used from the larger dataset. Eligible participants were required to complete a variety of online questionnaires (see section 3.5) to measure survivor's knowledge of cancer history and risk, core health beliefs (i.e., perceived vulnerability), anxiety, depression and HCU in the past two years. Participants

also self-reported demographic and clinical information (i.e., sex/gender, age at diagnosis, geographic location (rural/remote vs. urban) and annual household income (SES)). All questionnaires were administered online via Redcap, through a unique link provided to participants via email.

### **3.4 | Measures**

#### *3.4.1 Primary Outcome: Healthcare Utilization*

Our primary outcome measure and dependent variable of interest was HCU among YA SCC. The quantification of HCU among survivors was determined by asking participants to self-report the number of visits they had to a doctor in the past 2 years that were related to their initial cancer diagnosis. This two-part question was as follows: “During a 2-year period, how many times did you see a doctor?” and “How many of the visits to the doctor indicated in question #2 [see above] (during the 2-year period) were related to this previous illness [cancer]?” Survivors could select a quantity range of appointments as follows: 1) None; 2) 1-2 visits; 3) 3-4 visits; 4) 5-6 visits; 5) 7-10 visits; 6) 11-20 visits; and 7) over 20 visits.

#### *3.4.2 Sociodemographic Information*

Participants provided sociodemographic information, including but not limited to their age, sex (i.e., male or female), gender (i.e., male, female, or non-binary/trans-gender/two-spirit), race, SES (i.e., less than \$25,000; \$25,000-\$49,000; \$50,000-\$74,999; \$75,000-\$99,000; and more than \$100,000) and geographic location (i.e., rural/remote vs. urban). Participants were also asked to self-report all the types of HCPs that they had seen or talked to for medical care (routine or sick care) within the last 2 years and any current health problems that they perceived that they had in relation to their cancer diagnosis. Response choices for HCPs included: 1) none;

2) a primary care physician in the community (i.e., family physician); 3) a clinician at a cancer center (i.e., oncologist, nurse practitioner, other cancer specialist); 4) other medical specialists (i.e., endocrinologist, cardiologist, surgeon); 5) a psychiatrist; 6) a psychologist or counselor; 7) a physical or occupational therapist; and 8) other. Response choices for health problems included: 1) stroke; 2) problems with weaker bones; 3) lung problems; 4) having a different type or second cancer; 5) learning, memory, or attention problems; 6) cataracts; 7) dental problems; 8) fertility problems; 9) heart problems; 10) thyroid problems; 11) hearing problems; 12) mental health problems; 13) other; and 14) none.

### 3.4.3 Patient-Reported Outcomes Measurement Information System (PROMIS)

Participants completed the PROMIS SF v1.0-Anxiety 8a measure to assess anxiety score, as well as the PROMIS SF v1.0-Depression 8a to assess depression. PROMIS is a set of person-centered measures that can be used among children and adults within the general population, and those living with chronic disease (Cella D et al., 2010). These measures can evaluate and monitor various aspects of physical, mental, and social health. Each measure contains 8 items and asks participants to report on a Likert scale from 0 to 5, how often they experienced a specific feeling related to anxiety or depression, with 0 meaning never and 5 meaning always. Anxiety related questions included but were not limited to, “I felt fearful,” “I felt tense,” and “I felt anxious.” Similarly, depression questions included but were not limited to, “I felt worthless,” “I felt helpless,” “I felt depressed.” Raw scores are converted into T-scores with a mean of 50 and SD of 10. Higher scores represent more anxiety or depression. These measures have been used and validated within several different populations of survivors of childhood cancer (Patton et al., 2022) (Goode et al., 2022) and in the pediatric oncology setting (Hinds et al., 2013).

#### 3.4.4 *Self-Report Survey of Cancer Knowledge Questionnaire*

Survivor's knowledge of their cancer history and associated risks for LE was assessed using the *Self-Report Survey of Cancer Knowledge* questionnaire (Kunin-Batson., 2016). This is a 6-item survey designed to assess cancer knowledge. Included items were: 1) *Do you feel that your cancer treatments could cause serious future health problems?* 2) *Do you currently have a cancer survivorship care plan?* 3) *Did you receive chemotherapy?* 4) *Did you receive radiation therapy?* 5) *Did you receive a bone marrow transplant?* 6) *Did you receive any surgeries as part of your treatment?* Responses to items include: 1) No; 2) Yes; and 3) I don't know. For our study, Questions 1 and 2 were dichotomized as 1= Yes and 0=No/I don't know, and questions 3-6 were dichotomized as 1= yes/no and 0= I don't know. Total scores for each participant were obtained by summing all responses. Potential scores ranged from 0 to 6, where 0 represented inadequate knowledge of one's cancer history and risks, and 6 represented adequate knowledge.

#### 3.4.5 *Self-Report Survey of Core Health Beliefs*

A survivor's perceived vulnerability to risk of LE or secondary cancers was measured using 1-item from the *Self-Report Survey of Core Health Beliefs* (Tercyak et al., 2004) "How often do you wonder about cancer happening to you again someday in the future?". Responses to this item were rated on a 4-point Likert scale (1= not a lot, 4= a lot), where higher scores represented higher levels of perceived vulnerability.

### **3.5 | Statistical Analyses**

Demographic and clinical variables were described for our sample using means (SD) and frequencies. To address our subsequent aims, HCU was dichotomized based on the median

response (13.00, 3-4 visits), whereby HCU group 1, includes those who reported 0-2 visits, and HCU group 2, included those who reported 3+ visits.

To address aim 1, descriptive statistics were used to explore HCU among our sample. In addition, independent sample t-tests were used to identify the associations between patient-level factors and HCU, whereby the means of each variable was stratified by each HCU group (i.e., 0-2 visits, and 3+ visits). Finally, as a preliminary step to examining the associations between our independent variables and HCU, Spearman bivariate correlations were conducted. To examine these associations, race and SES were dichotomized prior to being entered into the final regression model. Race was categorized as either 0= non-white or 1= white/Caucasian. The dichotomization of SES was based on median (4.00, \$50,000-\$74,999), whereby 0= \$74,999 or less, and 1= \$75,000 or more annual household income (before tax).

To address aim 2, a multivariate logistic regression was utilized to investigate the association between HCU and patient-level factors. Logistic regression modeling is a statistical technique that describes and tests the relationships between a binary outcome variable (in this case, HCU) and one or more independent variables. Selection of independent variables for our analysis was determined based on our theoretical assumptions and by investigating the identified associations from aim 1. A backwards elimination process was used to build the elimination model whereby all independent variables were added to the model. To test the relevance of an independent variable on the overall results and fit of the model, one independent variable was removed at a time to see its effect. Odds ratios (OR) and 95% CI were reported as the output from the logistic regression model. SPSS version 29.0 software was used for all statistical analyses. A 2-tailed *p*-value less than or equal to 0.05 was considered statistically significant.

## **Chapter 4: Results**



## 4.1 | Participant Characteristics

In total, 280 survivors initiated this study, however, 123 survivors completed the study. Survivors were aged 18-39 years (mean=28.19, SD= 5.18) and were on average, 16.56 years post-treatment. 69.00% of survivors (n=85) reported that their assigned sex at birth was female. When asked to report their gender, 67.50% identified as female (n=83), 2.40% identified as being gender fluid, non-binary, or Two-Spirit (n=3), and the remaining 30.10% (n=37) identified as male. The most common cancer diagnoses were Leukemia (e.g., ALL, AML) (n=42, 34.1%), Lymphoma (e.g., Hodgkin's, Non-Hodgkin's) (n=25, 20.30%), Solid Tumor (e.g., Wilms tumor, Osteosarcoma) (n=25, 20.30%). The mean age of diagnosis among participants was 10.01 years of age (SD= 6.68). Survivors were from 7 provinces and 1 territory, predominantly Alberta (n= 45, 36.60%), British Columbia (n= 42, 34.10%), and Ontario (n= 22, 17.90%). The majority of survivors reported living in an urban setting (n=96, 80.0%). A detailed description of participant's demographic and clinical characteristics are outlined in Table 1.

**Table 1**

*Demographic and clinical characteristics of participants: young adult survivors of childhood cancer*

<b>Participant Demographic and Clinical Characteristics</b>	<b><i>n</i></b>	<b><i>%</i></b>	<b><i>M</i></b>	<b><i>SD</i></b>	<b><i>Median</i></b>
<b>Current Age</b>	119		28.19	5.18	
<b>Sex</b>					
Male	38	30.90			
Female	85	<b>69.10</b>			
<b>Gender</b>					
Man	37	30.10			
Woman	83	<b>67.50</b>			
Gender fluid, non-binary, or Two-Spirit	3	2.40			
<b>Age at Diagnosis (in years)</b>	123		10.01	6.68	
<b>Years Post Treatment</b>			16.56	7.54	

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**Diagnosis Type**

Leukemia (e.g., ALL, AML)	<b>42</b>	<b>34.10</b>
Lymphoma (e.g., Hodgkin's, non-Hodgkin's)	25	20.30
Brain Tumor (e.g., Medulloblastoma)	14	11.40
Solid Tumor (e.g., Wilms tumor, osteosarcoma)	25	20.30
Other	17	13.80

**Geographic Location**

Rural/Remote	24	19.50
Urban	<b>96</b>	<b>80.00</b>

**Province/Territory of Residence**

Alberta	<b>45</b>	<b>36.60</b>
British Columbia	42	34.10
New Brunswick	2	1.60
Nova Scotia	5	4.10
Ontario	22	17.90
Quebec	2	1.60
Saskatchewan	1	0.80
Yukon	1	0.80

**Relationship Status**

Single	<b>64</b>	<b>52.00</b>
Married	28	22.80
Living with a partner	28	22.80
Divorced	2	1.60
Prefer not to answer	1	0.80

**Education Level**

Graduate/Professional school, master's degree, PhD	17	13.80
Highschool	9	7.30
Some high school or less	5	4.10
Some university/college, no degree	25	20.30
University/College, bachelor's degree	<b>59</b>	<b>48.00</b>
Vocational school or training other than college	6	4.90
Prefer not to answer	1	0.80

**Race**

Aboriginal/First Nations/Inuit/Metis	5	4.10
Black/African/Caribbean	3	2.40
East Asian	16	13.00
Latin American	2	1.60
Middle Eastern	3	2.40
South Asian	7	5.70
White/European	<b>95</b>	<b>77.20</b>
<i>Other</i>	2	1.60

**Household Income (annual)**

Less than \$25,000	17	13.80
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\$25,000-\$49,999	10	8.10	
\$50,000-\$74,999	22	17.90	
\$75,000-\$99,000	22	17.90	
More than \$100,000	<b>29</b>	<b>23.60</b>	
Prefer not to answer	20	16.30	
<b>Religion</b>			
Buddhism	2	1.60	
Catholicism	15	12.20	
Christianity	30	24.40	
Islam	3	2.40	
Judaism	3	2.40	
Sikhism	1	0.80	
Other	<b>41</b>	<b>33.3</b>	
Prefer not to answer	27	22.20	
<b>Status in Canada</b>			
Citizen	<b>116</b>	<b>94.30</b>	
International Student	1	0.80	
Permanent Resident	2	1.60	
Other	1	0.80	
<b>HealthCare Utilization</b>			
None	23	18.70	
1-2 visits	<b>51</b>	<b>41.50</b>	
3-4 visits	25	20.30	
5-6 visits	13	10.60	
7-10 visits	3	2.40	<u>13.00</u>
11-20 visits	3	2.40	
More than 20 visits	5	4.1	<u>(3-4 visits)</u>
<b>Healthcare Providers</b>			
Primary care clinician in the community	<b>104</b>	<b>25.90</b>	
Clinician at a cancer center	95	23.70	
Psychiatrist	19	4.70	
Psychologist or Counselor	61	15.20	
Physical or Occupational Therapist	36	9.00	
Other medical specialist	73	18.20	
None	2	0.50	
Other	11	2.70	
<b>Health Problems</b>			
Stroke	2	1.60	
Problems with weaker bones	16	13.00	
Lung problems	12	9.80	
Having a different type or second cancer	8	6.50	
Learning, memory, or attention problems	50	40.70	
Cataracts	7	5.70	
Dental problems	15	12.20	
Fertility problems	32	26.00	
Heart problems	22	17.90	

Thyroid problems	25	20.30
Hearing problems	19	15.40
Mental health problems	<b>56</b>	<b>45.50</b>
Other	10	8.10
None	28	28.80

### **Aim 1:** Patient-level factors in relation to healthcare utilization

Our first aim set out to explore and describe healthcare utilization among our sample of YA SCC. Among our sample, reports of HCU ranged from no cancer-related visits within the past 2 years (n=23, 18.70%) to more than 20 cancer-related visits within the past 2 years (n=5, 4.10%). In addition, participants were asked to report the types of healthcare providers that they had seen or talked to over a 2-year period in relation to medical care for their cancer diagnosis. Only 0.5% (n=2) of participants reported that they had seen “none.” Both participants were in group 1 of HCU (0-2 visits). The three most utilized HCPs were primary care clinicians (n= 104, 25.90%), clinicians at a cancer center (n=95, 23.70%), and other medical specialists (n=73, 18.20%). Additional findings related to the frequency of HCP visits and HCP visits stratified by HCU groups are outlined in Table 1.0 and 2.0, respectively.

Participants were asked to self-report the health problems they think they currently have because of their cancer treatments. 45.50% (n=56) of respondents self-reported that they were currently experiencing mental health problems. Further, 40.70% of respondents reported that they had learning, memory, or attention problems. The third most common response was “none,” whereby 28.80% (n=28) of participants declared that they had no current health problems related to their cancer treatments. When stratified by HCU group, health problems were more commonly reported across all health problem types in HCU group 2, except for stroke, which was higher within group 1 (n=2, 0.50%). The most commonly reported health problems in group 1 included none (n=10, 8.10%); learning, memory, or attention problems (n=9, 7.30%); and mental health

problems (n=7, 5.70%). In group 2, mental health problems (n=49, 39.80%); learning, memory, or attention problems (n=41, 33.30%); and fertility problems (n=27, 22.00%) were the most reported health problems. Additional findings related to the frequency of health problems and health problems stratified by HCU groups are outlined in Table 1.0 and 2.0, respectively.

To further explore factors related to HCU, independent-sample t-tests were conducted to compare continuous variables and chi-square tests to compare categorical variables as a means of identifying the associations between each independent variable of interest (i.e., sex, gender, age, race, SES, geographic location, anxiety, depression, knowledge of cancer history, and perceived vulnerability), stratified by different healthcare utilization groups (i.e., 0-2 visits or 3+ visits). Results of independent-sample t-tests and chi-square analyses are described in Table 2.0. Anxiety ( $t(121) = -4.783, p < 0.001$ ), depression ( $t(121) = -2.874, p = 0.005$ ), and perceived vulnerability ( $t(121) = -3.216, p = 0.002$ ) were the only independent variables that had statistically significant differences in mean and standard deviation across HCU groups ( $p = 0.002$ )

**Table 2***Associations between patient-level factors, stratified by healthcare utilization groups*

		<b>HCU 0-2 Visits</b>		<b>HCU 3+ Visits</b>		P-Value
		N (%)	M (SD)	N (%)	M (SD)	
<b>Sex</b>	Female	17 (13.82)		68 (55.28)		0.090
	Male	13 (10.57)		25 (20.33)		
<b>Gender</b>	Female	17 (13.82)		66 (53.66)		0.137
	Male	13 (10.57)		24 (19.51)		
	Non-Binary, Gender-Fluid, Two-Spirit	0 (0.00)		3 (2.44)		
<b>Age</b>			29.30 (5.88)		27.87 (4.94)	0.210
	<b>Race<sup>1</sup></b>	White	9 (7.44)		17 (14.05)	
	Non-White	20 (16.53)		75 (78.51)		
		Aboriginal, First Nations, Inuit, Metis	1 (0.81)		4 (3.25)	
		Black, African, Caribbean	0 (0.00)		3 (2.44)	
		East Asian	9 (7.32)		7 (5.69)	
		Latin American	1 (0.81)		1 (0.81)	
		Middle Eastern	0 (0.00)		3 (2.44)	
		South Asian	2 (1.63)		5 (4.17)	
		Other	0 (0.00)		2 (1.63)	
<b>Annual Household Income (before tax) (SES)</b>	Less than \$25,000-\$74,999	11 (9.17)		38 (31.67)		0.829
	\$75,000 or more	18 (15.00)		53 (44.17)		
	Less than \$25,000	2 (1.63)		15 (12.20)		0.741
	\$25,000-\$49,999	2 (1.63)		8 (6.50)		
	\$50,000-\$74,999	7 (5.69)		15 (12.20)		
	\$75,000-\$99,000	4 (3.25)		18 (14.63)		
	More than \$100,000	9 (7.32)		20 (16.26)		
	Prefer not to answer	5 (4.07)		15 (12.20)		
<b>Healthcare Providers<sup>1</sup></b>	Primary Care Clinician in the Community	16 (3.99)		88 (21.95)		
	Clinician at a Cancer Center	17 (4.24)		78 (19.45)		
	Psychiatrist	1 (0.25)		18 (4.49)		
	Psychologist or Counselor	8 (2.00)		53 (13.22)		
	Physical or Occupational Therapist	2 (0.50)		34 (8.48)		

	Other Medical Specialist	9 (2.24)		64 (15.96)	
	None	2 (0.50)		0 (0.00)	
	Other	0 (0.00)		11 (2.74)	
<b>Health Problems<sup>1</sup></b>	Total Health Problems		1.40 (1.75)		2.49 (0.23)
	Stroke	2 (1.60)		0 (0.00)	0.016*
	Problems with weaker bones	2 (1.60)		14 (11.40)	
	Lung Problems	1 (0.80)		11 (11.80)	
	Having a different type or second cancer	1 (0.80)		7 (5.70)	
	Learning, memory, or attention problems	9 (7.30)		41 (33.30)	
	Cataracts	1 (0.80)		6 (4.90)	
	Dental problems	2 (1.60)		13 (10.60)	
	Fertility problems	5 (4.10)		27 (22.00)	
	Heart problems	3 (2.40)		19 (15.40)	
	Thyroid problems	5 (4.10)		20 (16.30)	
	Hearing problems	4 (3.30)		15 (12.20)	
	Mental health problems	7 (5.70)		49 (39.80)	
	Other	0 (0.00)		10 (8.10)	
	None	10 (8.10)		18 (14.60)	
<b>Geographic Location</b>	Remote/Rural	8 (6.67)		16 (13.33)	0.223
	Urban	21 (17.50)		75 (62.50)	0.241
<b>Anxiety</b>			49.09 (11.25)		59.65 (10.28)
<b>Depression</b>			49.19 (11.01)		55.71 (10.75)
<b>Knowledge of Cancer History &amp; Risks</b>			4.53 (0.63)		4.67 (0.56)
<b>Perceived Vulnerability</b>			2.03 (1.00)		2.73 (1.04)

Note

\*Significant at the 0.05 level (2-tailed)

\*\*Significant at the 0.01 level (2-tailed)

1: P-value not available as the variable was a multiple response question.

We further explored the associations among the independent variables and our primary outcome using Spearman's bivariate correlations (Table 3). Statistically significant positive correlations were found between HCU and anxiety ( $\rho = 0.364$ ,  $p < 0.001$ ) as well as HCU and depression ( $\rho = 0.249$ ,  $p = 0.005$ ), and HCU and perceived vulnerability ( $\rho = 0.281$ ,  $p = 0.002$ ).



**Table 3***Bivariate Correlations (Spearman's Rho) between Healthcare Utilization and Patient-Level Factors*

	Healthcare Utilization	Sex	Gender	Age	Race	SES	Geographic Location	Anxiety	Depression	Knowledge of Cancer History and Risk	Perceived Vulnerability
HealthCare Utilization	1.000	-0.153	0.176	-0.101	0.130	-0.033	0.107	0.364**	0.249**	0.102	0.281**
Sex	-0.153	1.000	-0.930**	0.070	-0.090	0.165	0.018	-0.058	0.030	-0.071	-0.177
Gender	0.176	-0.930**	1.000	-0.125	0.084	-0.162	0.004	0.043	-0.024	0.013	0.150
Age	-0.101	0.070	-0.125	1.000	0.052	0.090	-0.098	-0.146	-0.095	0.003	-0.159
Race	0.130	-0.090	0.084	0.052	1.000	-0.070	0.049	0.091	-0.010	0.044	0.131
SES	-0.033	0.165	-0.162	0.090	-0.070	1.000	0.091	-0.238**	0.173	-0.003	-0.100
Geographic Location	0.107	0.018	0.004	-0.098	0.049	0.091	1.000	-0.043	-0.107	0.058	0.247**
Anxiety	0.364**	-0.058	0.043	-0.146	0.091	-0.238**	-0.043	1.000	0.773**	0.231	0.374**
Depression	0.249**	0.030	-0.024	-0.095	-0.010	0.173	-0.107	0.773**	1.000	0.135	0.247**
Knowledge of Cancer History and Risk	0.102	-0.071	0.013	0.003	0.044	-0.003	0.058	0.231*	0.135	1.000	0.260**
Perceived Vulnerability	0.281**	-0.177	0.150	-0.159	0.131	-0.100	0.247**	0.374**	0.247**	0.260**	1.000

Note

\*Correlation is significant at the 0.05 level (2-tailed)

\*\*Correlation is significant at the 0.01 level (2-tailed)

## **Aim 2:** Relationships among patient-level factors with healthcare utilization

Our second aim set to examine the associations among demographic, clinical, and psychological patient-level factors with healthcare utilization among YA SCC. It was hypothesized that higher healthcare utilization will be associated with male sex and gender, rural/remote living, lower SES, older attained age, higher anxiety scores, and lower perceived vulnerability.

Variables selected for the logistic regression model were based on our theoretical assumptions and empirical evidence gleaned from Aim 1. As seen within table 3.0, strong collinearity existed between anxiety and depression ( $\rho=0.773$ ). Given the strong overlap between these two variables, we conducted two different models, where anxiety and depression were separated. Within Model 1, SES, race, geographic location, and sex were entered as categorical variables. The remaining independent variables, anxiety, knowledge of cancer history, and perceived vulnerability were entered as continuous variables. Similarly, SES, race, geographic location, and sex were entered as categorical variables in Model 2, and depression, knowledge of cancer history, and perceived vulnerability were entered as continuous variables.

Prior to conducting the regression, all assumptions were met. The dependent variable, HCU was binary in that it was classified as either 0: 0-2 visits, or 1: 3+ visits. All observations were independent of each other. While Bivariate correlation coefficient tests revealed that anxiety and depression were highly correlated ( $\rho=0.773$ ), depression was excluded from the model to avoid collinearity and meet the assumptions of the model. Outliers within the continuous variable Age were analyzed using Cook's Distance. All outliers were verified checked for legitimacy and were retained.

Model 1 was significant,  $X^2(8) = 23.68$ ,  $p = 0.003$ , and accounted for 29.3% of variance in healthcare utilization among YA SCC (Nagelkerke  $R^2 = 0.293$ ). 82.3% of cases were classified correctly. As seen in Table 4, anxiety was the only independent variable that added significantly to the model ( $p < 0.001$ ), and higher anxiety scores were associated with increased likelihood of higher healthcare utilization. Sex ( $p = 0.253$ ), age ( $p = 0.978$ ), race ( $p = 0.202$ ), SES ( $p = 0.315$ ), and geographic location ( $p = 0.548$ ) did not add significantly to the model.

**Table 4**

*Logistic regression examining associations among patient-level factors and HealthCare Utilization (Model 1)*

Independent Variable	B	SE	Odds	p	95% CI	
					Lower	Upper
Sex	-0.642	0.561	0.526	0.253	0.175	1.581
Age	0.001	0.052	1.001	0.978	0.904	1.109
Race	0.757	0.593	2.132	0.202	0.667	6.821
SES	0.567	0.564	1.763	0.315	0.584	5.324
Geographic Location	0.402	0.669	1.494	0.548	0.403	5.544
Anxiety	0.093	0.028	1.097	<0.001	1.038	1.160
Knowledge of Cancer History	-0.163	0.492	0.850	0.740	0.324	2.229
Perceived Vulnerability	0.262	0.285	1.300	0.358	0.743	2.273
Pseudo $R^2 = 0.29$						

Model 2 was significant,  $X^2(8) = 15.26$ ,  $p = 0.054$ , and accounted for 19.6% of variance in healthcare utilization among YA SCC (Nagelkerke  $R^2 = 0.196$ ). 77.0% of cases were classified correctly. As seen in Table 5, none of the independent variables added significantly to the model.

**Table 5**

*Logistic regression examining associations among patient-level factors and HealthCare Utilization (Model 2)*

<b>Independent Variable</b>	<b>B</b>	<b>SE</b>	<b>Odds</b>	<b>p</b>	<b>95% CI</b>	
					<b>Lower</b>	<b>Upper</b>
<b>Sex</b>	-0.598	0.545	0.550	0.272	0.189	1.077
<b>Age</b>	-0.023	0.050	0.977	0.643	0.886	1.109
<b>Race</b>	0.745	0.571	2.106	0.192	0.688	6.447
<b>SES</b>	0.391	0.534	1.479	0.464	0.519	4.214
<b>Geographic Location</b>	0.313	0.634	1.367	0.622	0.395	4.732
<b>Depression</b>	0.052	0.027	1.053	0.054	0.999	1.111
<b>Knowledge of Cancer History</b>	0.051	0.459	1.052	0.912	0.428	2.585
<b>Perceived Vulnerability</b>	0.433	0.271	1.542	0.110	0.906	2.625
Pseudo R <sup>2</sup> =0.20						

## **Chapter 5: Discussion**

Navigating a life after surviving childhood cancer is a lifelong and challenging journey for many YA. Both system and patient-level factors may create barriers to accessing timely and appropriate healthcare services for survivors. Such factors could result in the over or under utilization of healthcare services, meaning that some survivors might not be receiving the recommended care that they require to minimize their risks for LE and chronic conditions from their cancer treatments. Alternatively, some survivors may be contributing to excess burden on the healthcare system through overuse of healthcare resources. This thesis aimed to identify and explore the associations of survivor's demographics, knowledge of cancer history and risks, and psychological factors with HCU among YA SCC. Further, we aimed to examine the relationships among demographic, clinical, and psychological patient-level factors with HCU. The findings of this study may contribute to identified gaps in existing literature and help inform future studies that aim to improve appropriate use and access to HCU among SCC, while promoting health system sustainability to support the growing population of survivors in North America.

Our first aim planned to explore HCU among our population of YA SCC. Interestingly, a wide range of HCU rates were found within our sample. HCU rates ranged from no cancer-related visits within the past 2 years to more than 20+ cancer-related visits within the past 2 years. The median number of visits reported by participants was 3-4 visits. While the majority of our sample reported lower visit rates, it was interesting to see that almost 20% of our population reported more than 5 cancer-related visits. Data suggests that survivors who are 3 or more years post-treatment are typically seen once or twice annually (NIH, 2024). As the mean number of years post-treatment for our population was 16.56 years, and the median number of visits reported was 3-4 visits, our population is on par with the typical HCU rates seen among

survivors in current research. HCU rates that were on the extreme ends of the range (i.e., none and more than 20+) may have been due in part to participants interpretation of which appointments were cancer-related or if a singular visit with multiple components could be considered as multiple separate visits. The use of medical charts in future studies can help to verify that the reported number of visits is accurate based on the participant's history.

Other interesting observations from our exploration of HCU include the high prevalence of self-reported mental health problems (45.5%) among our sample, coupled with the limited use of psychiatrists, psychologists, and counselors. Cancer-related mental health difficulties are frequently cited in literature. Commonly cited mental health problems among SCC include but are not limited to anxiety and depression (Baclig et al., 2023), PTSS (Crochet et al., 2019; Baker & Syrjala, 2018), suicidal ideation (Baker & Syrjala, 2018), and cancer-related worry (McDonnell et al., 2021). Several factors may explain the high prevalence of mental health issues alongside the low utilization of mental health services found in our study. First, it is possible that there are unmet healthcare needs within our study sample. These findings are consistent with existing literature that emphasizes a lack of psychosocial and mental health supports as key themes in unmet needs reported by adult SCC (Hendriks et al., 2021; White et al., 2018). Alternatively, individuals with known mental health concerns may not be interested in seeking care or are already on a treatment plan for their mental health concerns. Some treatments for anxiety or depression may require a significant time commitment, which could limit the ability for some survivors to utilize these services. And finally, it is possible that system or knowledge barriers may be preventing some survivors from knowing which services are available, and or which they require. In consideration of the shortage of oncology workforce in Canada, including psychosocial support (Chaput, 2023), the use of digital health interventions

may be a valuable tool to promote self-management of anxiety and depression among YA SCC. Experimental evidence on the effectiveness of technology-based interventions for mental health management among SCC is still relatively mixed, however, a recent systematic review highlighted several instances of technology-based interventions having positive effects on anxiety, stress, and other psychological symptoms of cancer (Ozdimer Koyu & KilikarslanToruner, 2023).

Learning, memory, or attention problems were the second most reported health problems among participants (40.70%). It is interesting to consider how the role of learning or memory problems could negatively affect HCU. In considering the reliance on one's knowledge and memory to know which appointments are important based on risk and cancer history, knowing how to navigate the healthcare system, and remembering appointments, learning and memory problems may contribute to lower HCU rates, or the underutilization of services. No literature was identified that analyzed the role of cognitive problems and HCU among YA SCC. Given the high perceived prevalence of cognitive problems within our population, this may be an important area of research for future studies. The use of digital tools for appointment reminders and patient information-sharing may help improve access to healthcare services and appropriate risk-based HCU among YA SCC living with learning and memory challenges.

Consistent with our hypotheses, results of our study demonstrated that higher anxiety was associated with higher HCU, with a minimum of 3 cancer-related visits (as depicted by the HCU groups). This finding is consistent with findings from Crochet (2019) and McDonnell et al., (2021) where survivors experiencing PTSS, and cancer-related worry were more likely to have higher HCU rates specific to cancer-related care than those without PTSS or cancer-related worry. Several interesting points may be drawn from the findings of this current study related to



anxiety and HCU and the parallels from studies by Crochet (2019) and McDonnell (2021). First, given the interconnectedness of anxiety, worry, and PTSS with HCU, YA SCC are potentially using cancer-related healthcare services as a coping mechanism to deal with their cancer-related worry or anxiety through continuous testing and consultation with HCPs. It is also possible that frequent HCU perpetuates higher levels of anxiety, cancer-related worry, or PTSS among this population. This may be due in-part to distress or anxiety caused by difficulties in navigating the healthcare system or transitions from pediatric to adult care (Rosenberg-Yunger et al., 2012), or anxiety related to test results, sometimes referred to as “scanxiety” in the cancer context (Heathcote et al., 2022).

While little to no research exists that specifically studies the effects of anxiety on HCU among YA SCC, cancer-related worry has been found to be associated with anxiety in survivors of adult-onset cancer (McDonnell et al., 2021). A study of adult survivors of childhood cancer that studied associations between cancer-related worry and health behaviors (i.e., healthcare utilization) found that survivors who experienced cancer-related worry were more likely to have had 5 or more cancer-related physician visits and 5 or more calls to a physician over a 2-year period in comparison to survivors without cancer-related worry (McDonnell et al., 2021). While our findings related to anxiety and HCU are supported by existing research among adult SCC, further research is required that explores this association among a population of YA SCC.

The highly modifiable nature of anxiety with targeted behavioral interventions such as cognitive behavioral therapy (Zhang et al., 2022) and group therapy (Coughtrey et al., 2018) provide an opportunity to apply the findings of this study to help inform future interventions aimed at promoting risk appropriate HCU among YA. Interventions that are successful in lowering anxiety levels among survivors may not only contribute to the long-term well-being of

survivors through a decrease in psychological distress but may also perpetuate lower risk of LE among survivors through appropriate use of surveillance and testing services. Given the statistically significant associations between anxiety and HCU found within this study, interventions aimed at lowering anxiety levels among YA SCC could promote appropriate healthcare service use that improves the long-term QOL of survivors, and the long-term sustainability of the healthcare system.

In contrast to our hypotheses and existing literature, the current study did not find significant relationships with HCU and several variables including sex, knowledge of cancer history, and age. Non-significant findings between sex and HCU may be due in part to our study sample, that had a large majority of female participants. Although not explored in the current study due to limited variation with sex, it would be interesting to consider the role of gender in HCU, and other outcomes related to survivorship among YA SCC in future studies. As mentioned previously, sex and gender are often conflated in research and used interchangeably. The absence of research that analyzes gender as an independent variable of interest may contribute to challenges for future studies or interventions that primarily target gender differences and the overall generalizability of findings to a population of YA SCC.

Based on existing research related to age and HCU among SCC, the non-significant findings between age and HCU within this study raise concern, particularly for YA SCC for several reasons. Substantial amounts of evidence highlight the delayed onset of several late-effects and chronic conditions among SCC (Mertens et al., 2008; Armstrong et al., 2013; Turcotte et al., 2018). As such, if a survivor were to be receiving adequate risk-based care, their HCU rates should arguably increase as they age. Existing literature suggests that traditional aging challenges and complexities coupled with the risk of delayed onset of late effects among

SCC should be met with higher HCU rates to promote prevention and management of LE. Further concerns arise in relation to this finding in consideration of the typical long-term follow-up care structure in Canada for SCC. In Canadian long-term follow-up care clinics, it is common practice to reduce the frequency of follow-up appointments for survivors as the number of years post-treatment increases (Canadian Cancer Society, 2024<sup>A</sup>). However, the appropriateness of such practices should be reassessed considering research that emphasizes the delayed onset of chronic conditions and the heightened risk of late effects as survivors age (Mertens et al., 2008; Armstrong et al., 2013; Turcotte et al., 2018). Despite extensive research highlighted in the literature review emphasizing the importance of survivors having accurate and adequate knowledge of their cancer history and associated risks for proper healthcare utilization, total knowledge scores were not linked to healthcare utilization. Most of the study population were recruited from long-term survivor clinics, which may be contributing to these findings. Individuals who regularly attend follow-up care may be more likely to have higher levels of knowledge related to their cancer history and risks in comparison to those who were not actively pursuing any cancer-related follow-up care. To understand the associations between knowledge of cancer history and risks with HCU, future studies should aim to recruit individuals who are currently attending a long-term survivor clinic, and those who are not. This will provide a more accurate representation of the association between knowledge of cancer history and risk with HCU.

In contrast to our hypothesis, perceived vulnerability was not associated with HCU. There are several reasons that may explain this. While a survivor might recognize that they are vulnerable to LE and other co-morbidities related to their diagnosis, they may not choose to pursue health behaviors in relation to that perception. Drawing from the health behavior model,

Oeffinger's conceptual model of barriers and enablers highlights the interconnectedness between one's core health beliefs (i.e., perceived vulnerability) with their general motivations, cues to action, and self-efficacy. It could be hypothesized that if a survivor has higher levels of perceived vulnerability, but lower levels of motivation and self-efficacy, that they still might not utilize an appropriate level of healthcare services. Conversely, a survivor's low perceived vulnerability may be met with high levels of motivation and cues to action, resulting in overuse of healthcare services. It would be interesting to conduct a study that looks at the associations between one's core health beliefs (i.e., perceived vulnerability), external motivators (i.e., cues to action), and self-efficacy with HCU. Findings from Spearman's bivariate correlation analysis identified significant correlations between perceived vulnerability and anxiety.

Despite our hypothesis that those living in rural or remote areas would have lower rates of HCU, no statistically significant associations were found between geographic location and HCU. However, it is important to note that most of our study population were from urban settings, we did not have the statistical power to detect potential differences between urban and rural participants. Differences in the availability and accessibility of healthcare services in rural and remote populations in comparison to urban centers could contribute to significant differences in HCU rates among YA SCC. This is particularly important to consider for oncology care, whereby most cancer centers or long-term survivor clinics are situated in urban centers. This may prompt survivors from rural or remote settings to rely on primary care rather than specialized oncology care, demonstrating an important limitation of our primary recruitment strategies. Developing a strong understanding of geographic location and HCU among YA SCC is particularly important in the Canadian context, as 20% of the Canadian population is stated to live in a rural or remote area (Government of Canada, 2024). As such, future studies should

prioritize the recruitment of individuals from rural and remote settings to understand the association between geographic location and HCU among Canadian YA SCC.

## **5| Clinical and Research Implications**

There are several important clinical and research implications that can be drawn from the findings of this study. First, the identification of anxiety as a statistically significant predictor of HCU among our population of YA SCC could be targeted through behavioral interventions given the highly modifiable nature of anxiety. Such interventions may aim to reduce anxiety levels among this population which could improve survivor's overall psychological well-being, while potentially promoting appropriate risk based HCU rates and subsequent sustainability of the healthcare system. Findings demonstrating the substantial gaps between the number of perceived mental health problems among our population and the use of mental HCP services highlight potential unmet mental health needs for YA SCC. Given the consistency between these findings with existing literature, there is a strong foundation to support clinical improvements in mental health supports to help close the gap between mental health challenges and mental health support for this population. Non-significant findings identified between other patient-level variables and HCU were surprising but do not eliminate the need to consider such factors in the context of HCU. Finally, in consideration of the limited research that existed in relation to HCU among YA SCC in the Canadian context, the findings of this study provide a crucial basis for further research in this area. Ultimately, given the unique and complex experiences of YA SCC in combination with their associated high morbidity of LE and risk for chronic disease, the identification of patient-levels factors related to HCU within this study provide an important first step in ensuring that SCC can utilize appropriate risk-based care to help promote their long-term well-being.

## 9 | Strengths, Limitations & Future Directions

It is important to acknowledge both the strengths and limitations of this current study to help improve future work and provide context to the findings of this study. In acknowledging the limited availability of studies that analyze HCU among a population of YA SCC in the Canadian context, the findings of this study will contribute to an important, yet limited body of literature. Given the current lack of research surrounding the evaluation of patient-level factors associated to adherence to LTFU care amongst YA survivors of childhood cancer, this research has the potential to begin to close this research gap, while informing future studies relating to health behavior interventions within this population. Further strengths reside in the quantitative research methods scientifically supported for this specific research area, increasing the accuracy and reliability of results. Finally, long-lasting partnerships that we have developed with our patient partners have helped to inform our research and align our findings with the perspectives and needs of this population.

A primary weakness of this study is the small sample size and the limited diversity within our sample. Both factors limit the generalizability of our findings. Limited diversity existed across several participant demographic characteristics, including sex, geographic location, and race. Given that 69% of our sample were female, this may have weakened the validity of findings between sex and HCU. Similarly, diversity in our sample's race was limited, where the majority of participants were white. As such, the validity of the associations between race and HCU may have been weakened, and the generalizability of these preliminary findings to a non-white population are not appropriate. Further weakness resounds in the fact that this study did not utilize medical history records to verify patient information related to HCU. As such, it is possible that when self-reporting the number of visits that participant had to a doctor in the past

2-years related to their diagnosis, they could have improperly recalled the correct number categorized specific long-term follow-up care procedures as additional appointments. This could have resulted in the underreporting or overreporting of healthcare utilization. Not having access to medical records also meant that we were unable to determine if the number of healthcare visits were required or prescribed by a treating physician or whether they were at the request of the survivor.

Primary recruitment of participants from the Long-Term Survivor Clinic at the Alberta Children's Hospital, and other regional long-term follow-up clinics may have contributed to a higher proportion of our population being in the higher HCU group given their current involvement in their cancer-related care. Similarly, social media recruitment may have gathered a similar population of individuals given that they are involved on social media with cancer research accounts. The presence of anxiety may have contributed to selection bias within our sample, whereby survivors with higher levels of anxiety may have been more likely to have been attending a survivor clinic that was involved within recruitment. Alternatively, anxiety levels may have acted as a barrier to recruitment if those who exhibited higher levels of anxiety intentionally avoided healthcare or research settings.

While not being addressed within the context of this study, it is important to highlight that a large portion of identified research among SCC analyzed different diagnosis types and treatment modalities (i.e., radiation versus chemotherapy) as one homogenous group, which may create discrepancies in the generalizability of HCU findings and future areas of intervention. The separation of different diagnosis groups to understand HCU is important given differences in the long-term risks associated with different treatment modalities and diagnosis characteristics (i.e.,

severity, location, etc.) and should be further explored within a study that is able to verify diagnostic and clinical information from patient medical records.

The findings of this study provide a valuable first step in understanding the relationship between patient-level factors, demographics, and their relationship with HCU among YA SCC. Future work utilizing the findings of this study should aim to 1) verify findings through an analysis among a large, diverse, and representative population of YA SCC; 2) utilize medical records to understand differences in perceived and actual health complications, as well as the types and quantity of reported HCU visits; and 3) recruit additional individuals who are no longer attending long-term follow-up care.



## **Chapter 6: Conclusion**

The results of this study revealed a wide range of HCU among our survivors and showed that anxiety was a key factor related to greater HCU. The highly modifiable nature of anxiety using behavioral interventions provides promise towards improving the psychological well-being of survivors and promoting the use of risk appropriate HCU among this population. Given the high morbidity of LE and risk of chronic health conditions among YA SCC and the importance of appropriate HCU for the long-term well-being of survivors, the findings of this study are worthy of continued investigation.

The interconnectedness between the well-being of an individual and the well-being of the health system that sustains the individual is often overlooked. It could be stated that a sum is only as good as its parts. This is particularly true for survivors of childhood cancer and the Canadian healthcare system. To ensure the long-term well-being of survivors, improvements in childhood cancer survival rates must be met with improvements in the quality and accessibility of survivorship care. While a survivorship journey is often a long and windy road for many SCC, commitment to improvements in clinical care and research can help pave their path.

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