IMPROVING THE TRANSITION FROM PEDIATRIC TO ADULT CARE FOR SURVIVORS OF PEDIATRIC CANCER IN NEWFOUNDLAND, CANADA

by © Devonne Ryan

A thesis submitted
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Abstract

Advances in the treatment of childhood cancer have led to improved survival rates. Yet, childhood cancer survivors (CCS) have greater health risks and other health burdens that require appropriate follow-up and surveillance throughout their lives. Higher survival rates mean that significantly more pediatric oncology patients will reach adulthood and will be transitioned into adult care for their continued follow-up or aftercare. The transition from pediatric to adult-centered care is, however, increasingly being recognized as a period during which patients risk become disconnected with the healthcare system and having poorer health outcomes. This situation can be potentially very troubling for patients who require continued, regular surveillance. The focus of this doctoral research is to explore the transition from pediatric care to adult care and aftercare practices for CCS in Newfoundland (NL), Canada.

Guided by the World Health Organization's *Quality of Care: A Process for Making Strategic Choices in Health Systems* framework, we first reviewed models of care (MOC) and interventions aimed at improving aftercare that had been evaluated in the academic literature, including those specific to transition. We then conducted a qualitative study to detail the processes and barriers of transition for CCS in NL. Based on this information and discussions with local stakeholders, we identified the need for locally relevant educational resources. We then developed an educational workbook, titled *After the Janeway*, to be used by CCS in NL during their transition into adult-focused aftercare. Finally, we evaluated the workbook using a validated scale for assessing patient educational materials. The assessments clearly show that the adoption of this workbook into clinical practice should improve the experiences of CCS transitioning into adult care in NL. Collectively, our studies establish a thesis that is novel and potentially serves as the groundwork for future research related to improving the quality of the transitions and aftercare for CCS.

Dedication

This dissertation is dedicated to my father, the late Ron Ryan. My father embodied passion in everything he did. This dissertation would not be possible without hard work, tenacity and accountability, all traits modeled by my father. He taught me that when the “goings get tough, the tough get going” -- a lesson that I’ve held close throughout my graduate studies. When I told my father, I would be starting a doctorate, he said, “Great! But you better finish it.” Here you have it dad, a finished Ph.D!

Acknowledgements

It is my pleasure to recognize everyone who played an influential role throughout my doctoral program. The doctoral process has been a humbling experience and one in which I have been challenged by my own weaknesses and needed to learn to lean on others for support. For everyone I leaned on over the past few years, I hope I didn’t knock you over. Thank you and I hope you too feel a weight lifted!

My family has been formative in my educational endeavors. A special thank you to my father, Ron Ryan. I am forever indebted for all you have given me. You inspired me to push myself beyond what I thought I was capable of and reminded me to always hold myself accountable to my efforts. I promise that your energy, hard-work, determination and commitment were carried through this dissertation and will continue to be cherished throughout my future undertakings. To my mother, Rosalie, I can’t say enough about your endless love and support. Thank you for always giving me someone to talk with and for keeping me energized with amazing home cooked meals. I am beyond fortunate to have you in my life. To my sister, Natalie, you’ve been my rock. Despite living thousands of miles away, you’ve been by my side each step of the way. I’m appreciative of your continuous support, frequent visits, and all the meals you made me (especially the three months’ supply as I prepared for my comprehensive exams). Your humor carried me through these past 5 years and I am forever grateful to have you in my life. Thank you for celebrating each milestone with me and always being willing to adventure along-side me. To my brother, Ronald, your patience is appreciated. Thank you for putting up with my selfishness over the last few years. To my partner, Adam, I've been fortunate to have you one step ahead in the Ph.D. process. Feeling the passion, you bring to your field of study inspired me in moments when I lacked motivation. Thank you for reminding me that adversity does not need to be a stumbling block—but rather a stepping stone to something great. Seeing you persevere when faced with enormous personal and professional challenges, helped remind me in difficult times how to be appreciative for all that I have. While as frustrating as it can be, I thank you for always challenging my ideas. My ability to logically and critically assess information is much better because of your stubbornness. Thank you for travelling the world alongside me—even when you didn't want to. And most importantly, thank you for your "punny" sense of humor. I wouldn't have made it this far without you. Your calmness and patience with me have been extraordinary. Your fight for life is inspirational!

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List of Abbreviations

Acute Lymphoblastic Leukemia (ALL)

Adult Follow-up Survivorship Clinic (AFSC)

American Academy of Pediatrics (AAP)

Canadian Pediatric Society (CPS)

Centers for Disease Control (CDC)

Childhood Cancer Survivor(s) (CCS)

Childhood Cancer Survivor Study (CCSS)

Children's Oncology Group (COG)

Consolidated Criteria for Reporting Qualitative Research (COREQ)

Healthcare Provider (s) (HCP)

Long-term Follow-up (LTFU)

Model of Care/Aftercare (MOC)

Newfoundland and Labrador (NL)

Physical Activity (PA)

Pediatric Oncology Group of Ontario (POGO)

Post-Traumatic Stress Disorder (PTSD)

Preferred Reporting Items for Systematic Reviews & Meta-Analysis (PRISMA)

Primary Care Provider (PCP)

Quality of Life (QOL)

Randomized Controlled Trial (RCT)

Strengthening the Reporting of Observational studies in Epidemiology (STROBE)

Success Through Education, Psychosocial support, and Socialization Program (STEPS)

Surviving Cancer Competently Intervention Program (SCCIP)

The Patient Education Materials Assessment Tool for Printable Material (PEMAT-P)

World Health Organization (WHO)

List of Appendices

List of Publications

The following publications have been derived from the work of this dissertation:

Ryan D, Chafe R, Hodgkinson K, Chan K, Stringer K, Moorehead PC. Interventions to improve the aftercare of survivors of childhood cancer: a systematic review. Pediatric Hematology Oncology Journal. 2018;3(4):90-8.

Ryan D, Moorehead PC, Chafe R. Standardizing the categorizations of models of aftercare for survivors of childhood cancer. BMC Health Services Research. 2019;19(1):1-6.

Ryan D, Moorehead PC, Chafe R. Evaluating a Transition Workbook for Childhood Cancer Survivors: a Pilot Study. Journal of Cancer Education. 2020; (26):1-6.

Ryan D, Chafe R, Moorehead P. Transition from pediatric to adult aftercare for survivors of pediatric cancer in Newfoundland and Labrador: a qualitative study. CMAJ open. 2021 Apr;9(2):E309.

Drafted for Publication

Ryan D, Chafe R., Moorehead PC, Hodgkinson K, Chan K, Stringer K. Models of care for childhood cancer survivors once they become adults: A Systematic Review.

Chapter 1

Introduction

Background

Due to advances in treatment, the 5-year survival rate for children who get cancer is now over 80% (1). This impressive gain in survival means that significantly more pediatric oncology patients will reach adulthood and, ultimately, will be transferred into adult care (2). While the dramatic improvement in the prognosis for childhood cancers is encouraging, childhood cancer survivors (CCS) still face significant health risks (1, 3, 4) and health system challenges (5, 6). Adult survivors of childhood cancers are at higher risk compared to their peers of developing subsequent cancers (7), of having physical co-morbidities associated with all body systems (8), increased body weight (9), educational or neurocognitive impairments (10), mental health issues, fertility concerns (11) and even complications due to medical posttraumatic stress (12, 13). In fact, it is estimated that approximately 2/3 of CCS will be in some way negatively impacted from their previous cancer and by late effects of its treatment. This wide range of health risks and burdens mean patients require appropriate follow-up and surveillance throughout their lives (2, 14).

Figure 1.1 illustrates the disease-treatment progression for survivors of childhood cancer (Original Figure). The top row shows the disease progression, while the bottom row highlights the different phases of treatment. The disease progression includes the onset of the disease, recognition of symptoms, treatment and the ongoing risk of reoccurrence and late effects. Both the ongoing risk of reoccurrence and late effects for a CCS start while patients are still in pediatric care and continue into adulthood. The progression of care includes diagnosis, active treatment, pediatric aftercare, transition into adult aftercare and adult aftercare. If there is a secondary cancer, this cycle would repeat. For many patients, as they approach adulthood, their pediatric aftercare ends and they are transitioned into the adult system to receive aftercare (6). This project focuses on both the transition from child-centered aftercare and subsequent adult-centered aftercare in hopes of improving the experience and outcomes of CCS during early adulthood.

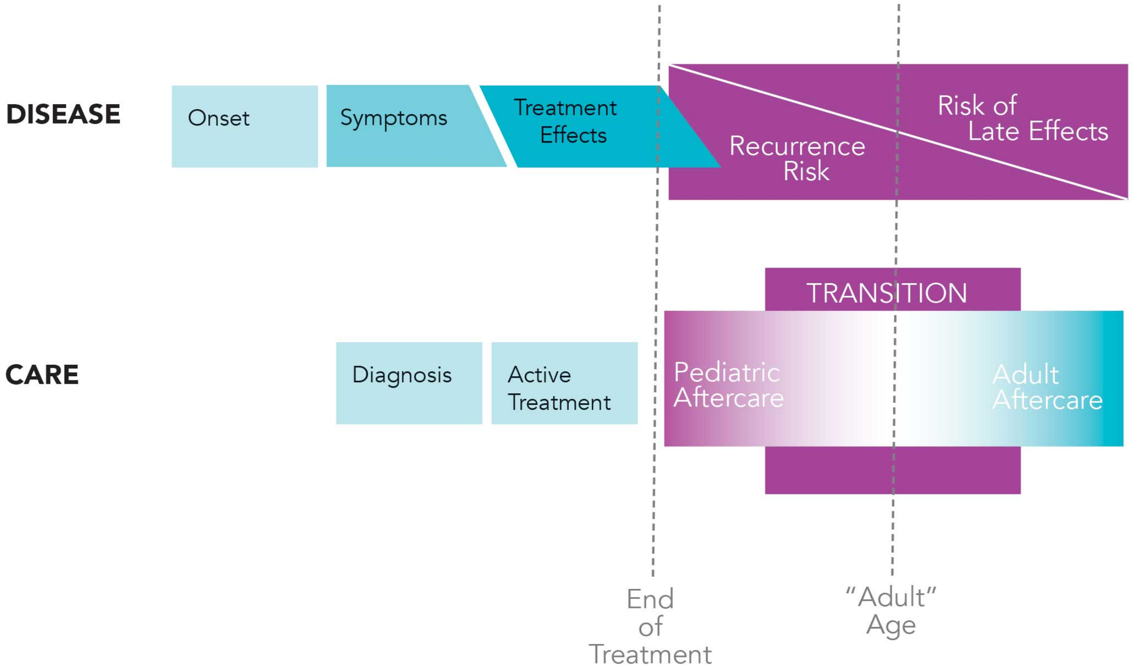

Figure 1.1 *Disease-Treatment Progression for Childhood Cancer Survivors (Original Figure)*

Aftercare is the care a CCS receives following the completion of their cancer treatment (15-17). It should provide CCS with regular contact with a healthcare team that is trained to understand cancer, cancer treatments, and the potential for long-term or delayed side effects of cancer treatments, i.e., late effects (18). Aftercare should focus on the identification and treatment for any problem that may arise as a result of cancer treatment, including any referrals that are needed in order to address issues when they arise (2). It should also include educational resources for CCS. Education is particularly important during the transition to adulthood, when CCS become increasingly responsible for their own healthcare needs.

The transition from adolescence to adulthood is a major developmental milestone for any individual (19). Individuals are at a vulnerable stage in their life, where they are faced with a great deal of growth and change. As adolescents move into adulthood, they are developing their self-identities and becoming increasingly independent. They may be living without their parents or guardians, attending post-secondary education, entering the workforce and developing different relationships. This complex time in a person's life is further complicated by a past cancer diagnosis. It is during this period of developmental growth that CCS are usually required to make the transition from the supported system of pediatric care into the adult system where they are often expected to take more responsibility their own care. Further, it is expected that CCS have knowledge of their healthcare issues and be more involved in their disease management if there are chronic complications (6, 19).

Transition to adult care is supposed to be a "purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centered to adult-oriented health-care systems (20)." The transition should be conducted gradually and start in the pediatric system. Further, transition should be clearly communicated between the patient, family, and HCP (21). Unfortunately, many CCS do not receive a formal or structured transition from their pediatric provider into adult care (22). In turn, many CCS do not continue attending aftercare as young adults. Inadequacies in transition leave CCS more vulnerable to not receiving proper care later in life. Studies have also shown that CCS are not always well informed about the type of cancer they had or its treatment, which can further hamper their ability to seek out subsequent care if it is needed (5, 14, 22-26).

Improving transition is not simply a clinical issue concerning linking patients to the right care provider. It is also a health systems and policy issue that involves ensuring improved system integration to support good follow-up and developmentally appropriate, patient-centered care (6). One approach for identifying health system gaps and improving care is the *World Health Organization's* (WHO) *Quality of Care: A Process for making Strategic Choices in Health Systems* (details presented in Chapter 2). This framework aims to bring about change in the healthcare system through a structured approach, focused on the available evidence and a local context. With a shift in emphasis across the Canadian healthcare system toward more patient-centered care and improved integration between the specialist and primary care systems, the need to improve transition of CCS into adult care has never been more urgent (27).

In this thesis project, I use the WHO framework to identify ways to improve the transition from pediatric to adult care for CCS.

Rationale for Dissertation Research

We know that CCS face the complexities of their disease while they transition from pediatric care to adult care (6). This transition can be challenging because so many things are changing during this period in an individual's life. CCS are confronted with the challenges of adhering to recommended aftercare practices, such as continued screening and surveillance over the course of their lives. With huge improvements in CCS survival rates come new challenges to improve evolving and, at times, challenging realities for CCS and their families. Given the far-reaching impacts for CCS, it is imperative that HCP can ensure that transitioning to adult care is a seamless practice and that CCS understand the critical importance of their aftercare (6, 19).

The evidence is clear that CCS are at risk of developing late effects; what is not clear is what the best approach for structuring and supporting aftercare for this population. In Canada, only 71% of pediatric centers have a dedicated program or clinic for survivors in both pediatric and adult care (28). This thesis project is practical in focus and is directly aimed at changing care in the field of cancer survivorship.

The transition from pediatric to adult-centered care is increasingly being recognized as an important issue and one tied closely with aftercare (6, 19). This transition occurs naturally for those without major health complications in childhood; however, for CCS, it serves as a point of stress. When transition fails to meet the requirements of CCS, they can "fall through the cracks" and be faced with a deterioration of their health or disconnection with the healthcare system – both having detrimental effects on their long-term health (6, 19, 21). The importance of aftercare is well documented, yet many healthcare centers do not have a transition program in place to ensure a successful transition into adult aftercare (13). Unfortunately, transition has long been seen as a handoff or transfer to the adult system (6, 26). In fact, in Canada only 12% of pediatric centers offer transition programs (28). Living with and managing lifelong potential late effects requires adjustment on many levels to achieve quality transition care that leads to successful aftercare. Therefore, a clear understanding of this process is needed. We must: (a) understand the current aftercare practices for CCS, (b) determine the current transition processes in place for CCS, (c) identify barriers to and facilitators of successful transition, and (d) examine effective interventions to improve practices.

Effectively understanding the transition and aftercare processes is critical for ensuring good health outcomes for this population. The purpose of this research project is to understand the literature and the local context in order to develop an intervention to improve aftercare for CCS who were initially treated at the Janeway Children's Health and Rehabilitation Centre (St. John's, Newfoundland and Labrador (NL)), which is colloquially referred to as the Janeway: a term I will use from this point forward. We chose this population because it is unique in many aspects. NL is a defined geographic region and has only one pediatric cancer program that treats both urban and rural patients. Anecdotally, the retention rates at the Janeway are considered high for the pediatric aftercare clinic. As the Janeway is the only hospital that cares for pediatric oncology cases in NL, we have a unique opportunity to use our findings and apply a suitable intervention to the entire provincial population.

Research Paradigm

Reflexivity

Reflexivity is regularly considered as the process of reflecting critically on oneself as a researcher (29). Aspects of reflexivity may include ongoing internal dialogue and self-evaluation of the researchers' position. Reflexivity may also include acknowledging any position the researcher may be in that could affect any processes and outcomes of the research (29, 30). Reflexivity plays a pivotal role in qualitative research. Understanding how a researcher creates knowledge, monitors their biases, and understands their beliefs all play a role in the research process. Reflexivity allows the researcher to involved in the process as well as the product of research and is a means to enhance the rigor of a study (31). As such, it is important for me to present my own position as the researcher.

My interest in the field of aftercare was founded long before I entered into the academic world. As a young adult, I watched a close family member slip through the cracks between pediatric and adult care. This left a lasting understanding of the importance of continued care through health systems regardless of disease. Albeit, it was never a path I considered pursuing academically. That is, until one day, in a serendipitous fashion, a research proposal about *after care* was presented to me. My ideas around this research study manifested over the next few days. When I began my doctoral research, I, like all researchers, was confronted with the question, “what is the best way to investigate my research problem?” My experiences became the trigger for me to ask questions that guided my research.

One position that should be considered includes how my own experiences shaped how I engaged and developed relationships with participants of this research. I believe having an outside experience with issues related to transition allowed me to develop a deeper investment and passion for the field. Acknowledging my relationship to the field early in the process helped to recognize the potential impacts on the interpretation of the data, such as my own understanding of the importance of transition in healthcare.

Theoretical Paradigm

A theoretical paradigm is a "set of basic beliefs that deals with ultimate's or first principals (32)." It represents a view that defines a researchers' view on the world and how they are situated within it (32).

The guiding theoretical paradigm of this research is pragmatism. Pragmatism is best suited for the mixed-methods design because it allows for the researcher to choose the data collection methods as they pertain to the issue under investigation. This paradigm is suited for the sequential nature of this research and allows for both open- and closed-ended questions found in both qualitative and quantitative data and analysis (33, 34). My research presents a sequential process, guided by the WHO framework, for improving transition from pediatric to adult care for CCS and makes use of both qualitative and quantitative methods. Given the nature of this area of study, pragmatism allows for the use of method and philosophy to fit together. Pragmatic knowledge allows for the development of knowledge through integration of data at various stages of inquiry. Pragmatism allows for the greatest opportunity to answer the outlined researcher objectives and questions of this dissertation.

This research project availed of the mixed methods research design. Mixed methods research is common to research practice and recognized as the third major research approach, along with qualitative research and quantitative research (35). The use of mixed methods for this research project allowed for both open and closed-ended questions, multiple forms of data, and statistical and text analysis. This research design allowed us to consider multiple perspectives in our attempt to improve aftercare for CCS.

As is common with mix-methods studies, results from one method are used to inform other methods (35). This mixed-method study was conducted using sequential procedures. We used the systematic review articles to inform our qualitative study. The qualitative study was used for exploratory purposes. This was followed by a quantitative study that honed in of areas needing improvement identified in the previous study. Triangulation of the data was met by using multiple participants, data collections, and study designs (36). The WHO Framework was used in parallel with the research paradigm to assess, plan, implement and evaluate as per the study research objectives. This allowed for convergence across the mixed methods with the hope of developing a quality improvement intervention that was both context-appropriate and effective.

Research Objectives

The central research question addressed in this dissertation is: *How can aftercare for childhood cancer survivors be improved in NL?*

The research objectives for the project are:

1. To review the models of aftercare (MOC) and interventions for aftercare of CCS currently described in the academic literature, including those relating to transition;
2. To develop a detailed description of the processes by which young adult CCS transition from pediatric care to adult care in NL;
3. To examine CCS's and their families' experience of transition to adult care in NL;
4. To identify limitations, gaps in services and barriers to improved care for CCS in NL;
5. To identify, develop, and evaluate an intervention aimed at improving transition for CCS; and
6. To disseminate findings that may have policy implications for better supporting CCS to key stakeholders.

Program of Research for Dissertation

This section details my (Devonne Ryan) substantial contribution to this program of research and clarifies the roles at the level of the individual and that of the team.

This research included two systematic reviews. One of these reviews focused on models of care for CCS and the other focused on interventions to improve aftercare. Upon completion of these reviews, a gap was found in the literature: transitions for CCS were vastly understudied. A case study was developed to understand the practice of transition into adult aftercare at the Janeway. Based on this information and discussions with local stakeholders, the need for locally relevant educational resources was identified. An intervention was then developed and evaluated to improve the transition from pediatric care to adult care for CCS.

A project proposal was developed, and funding was secured for the project prior my joining the team. Upon joining the research team in 2014 I was fundamentally involved in the management and conduct of all aspects of the project and took the lead in: (a) developing the review protocol and completing a systematic review of models of care for CCS; (b) developing the review protocol and completing a systematic review of interventions for CCS; (c) completing and submitting a research ethics application for the project; (d) planning, recruiting participants, collecting data and analyzing the case study research; (e) assessing the research to determine the need for an effective transition specific intervention for CCS; (f) creating the workbook intervention *"Life After the Janeway;"* (g) evaluating the intervention using the PEMAT-P (The Patient Education Materials Assessment Tool for Printable Material) survey; (h) writing and submitting articles for publication; and (i) writing this dissertation. I am first author on all the manuscripts included in this dissertation. This dissertation is my independent scholarly work. However, throughout this thesis I will sometimes use the terms "the research team" and "we" to reflect the collaborative efforts of the research team.

Manuscript Format

This dissertation is presented in a manuscript format. An introductory chapter, literature review, and a closing chapter are the bookends to the five manuscripts (presented in Chapters 3-5). Chapter 2 presents a general literature review and the framework that the research is based on. Chapter 3 presents the findings of a systematic review of both models of care and interventions for CCS. It also includes an article that outlines our categorization of the different models of care. Chapter 4 presents a case study of HCP and CCS as they transition from pediatric care adult care in NL. Chapter 5 summarizes an intervention to improve aftercare for CCS in NL and presents its evaluation. Finally, Chapter 6 includes a general discussion of the findings, the implications for clinical practice and future research, and conclusions.

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Chapter 2

Literature Review

Introduction

In this chapter, I will provide relevant background information on the prevalence and survival rates of childhood cancer, late effects, aftercare for CCS, the transition from pediatric to adult care, MOC and interventions for aftercare. This information will help provide a wider context for the two systematic reviews that are presented in the next chapter. I will also discuss the WHO's *Quality of Care: A Process for making Strategic Choices in Health Systems* - the framework chosen to help structure my overall research project.

Childhood Cancer Statistics

Globally, an estimated 250,000 children and adolescents each year are diagnosed with cancer (1). In Canada, there are approximately 2075 new cases of cancer diagnosed each year in adolescents and young adults between the ages of 15-29 years, and 836 new cases between the ages of 0-14 years (2). The most common pediatric cancers for children ages 0-14 years include leukemia, lymphoma and brain/central nervous system cancer (3). For those between 15-29 years of age, the most common forms of cancer are thyroid, testicular, Hodgkin lymphoma and melanoma (4). The five-year survival rate for all pediatric cancer has increased considerably over the past four decades, now reaching over 80% (5).

Childhood Cancer Survivors and the Risk of Late Effects

Cancer treatment often requires aggressive therapies, such as chemotherapy or radiation therapy, to help patients overcome the disease (7-9). These therapies have the potential to cause harm to various organs and tissues, which can present as health issues later in life (10). Late effects are conditions that cancer survivors can have after they have completed their cancer treatment. According to the *National Cancer Institute* (U.S.), a late effect is "a health problem that occurs months or years after a disease is diagnosed or after treatment has ended (6)." They are common in all systems in the body, as well as potentially impacting psychosocial functioning. Some CCS can also suffer from post-traumatic stress disorder (PTSD) from their condition and its treatment (3).

Due in large part to the high survival rates of pediatric cancers, the impacts of late effects are increasingly being recognized as important aspects of a patient's cancer experience. In fact, between 60 and 70% of CCS are at risk of developing late effects due to their disease and treatment (11). Late effects and risks are different for CCS, as compared to survivors of cancers treated in adulthood, because they received their treatment early in life, when they are still developing. Examples of this include the severe toxicity associated with central nervous system irradiation in very young children or effects of treatment on growth (12).

Aftercare for Childhood Cancer Survivors

Aftercare is the follow-up care CCS receive when their treatment for cancer is complete (19-21). For CCS, aftercare starts in pediatric care institutions and continues into adult care (See Figure 1.1). Some institutions have aftercare clinics that specialize in aftercare for CCS while others will transfer CCS to primary care providers (PCP) or clinics (2,8,16,20,21). Taking part in aftercare allows CCS to remain in control of their health. Without aftercare, poorly managed late effects can have long-lasting consequences for CCS (13, 14). As a result, some pediatric oncology centers have aftercare programs that recognize and intervene early in an effort to reduce adverse late effects (15-17). There are aftercare guidelines for HCP to follow; however, these guidelines are generally not well adhered to (22). Yan et al. conclude that guideline adherence is lacking and that new strategies to improve adherence are necessary (22). Studies also show that it is a fundamental challenge to provide continued aftercare to CCS into adulthood (19). CCS face unique vulnerabilities related to a number of variables outside the cancer diagnosis and treatment. Several of these variables include: the coordination of care, social support network, knowledge and education of cancer, location, comprehensive care (8, 19-21).

In Canada, most CCS are treated at one of seventeen pediatric cancer programs (23). A 2009 study by Ristovski-Slijepcevie et al. showed that only 71% of programs had a formal aftercare program even though 88% reportedly adhered to the LTFU guidelines for CCS. Further, 76% of pediatric centers provided a treatment summary or a survivorship care plan before CCS transferred out of the pediatric program. However, only one center reported having a formal transition program to support CCS move from pediatric aftercare to adult aftercare (23).

There are some gaps and issues that can negatively impact a patient's aftercare. CCS, their families, and HCP should understand the structure and value of aftercare programs. It is also important that CCS understand their past treatments and the potential health risks that are associated with them. Unfortunately, evidence suggests that only 30% of CCS understand their risk for late effects (3). Aftercare programs should meet the medical needs of CCS to minimize potential late effects (3). HCP have specific guidelines to help monitor late effects. For example, the Children's Oncology Group (COG) Long-Term Follow-Up (LTFU) Guidelines for Survivors of Childhood, Adolescent, and Young Adult Cancers is a resource for healthcare professionals caring for this population (18). If CCS do not receive continuity of care of late effects, they may be less likely to successfully transition into adult aftercare and continue to monitor their health (2).

Models of Care for Childhood Cancer Survivors

A MOC refers to the structure of the aftercare services delivered to CCS. There is a variety of models for delivering aftercare to CCS, with each MOC having their own set of advantages and disadvantages. While some models of care are carefully planned and designed, other MOC may be default models where minimal access to care is available. The model best suited for an institution is dependent on the population of

CCS and the resources available in that facility (3, 24-26). CCS have unique needs that can be medical, educational and psychosocial in nature. A good MOC should seek to address all these aspects of care (3, 8, 24-27). It should also be stated that not all institutions have a MOC and/or program for CCS to transition into. *Table 2.1* highlights some of the different types of models of care for CCS. More details on models of care are described in Chapter 3, which presents a completed systematic review of models of care for CCS.

Table 2.1 *MOC and Description*

Aftercare	**Description**
Pediatric-Led Model	A pediatric oncologist and/or pediatric oncology program is primarily responsible for providing programs and are located in a pediatric hospital (23, 28).
PCP-led model	The community-based follow-up is being conducted mostly by a PCP often without support from oncologists. This model is generally outside a hospital setting.
Hybrid Oncology/Primary Care Model	PCP are responsible for monitoring patients' late effects but CCS also continue to have a connection with a cancer physician and/or program.
Adult Oncology-Led Model	An adult oncology team is the primary point of contact. This can be in the form of appointments or clinics. This model is generally inside a hospital setting.
Nurse-Led Model	Nurses are responsible for a variety of aftercare services. Nurses are the primary point of contact and the services may include medical follow up to providing counseling and education about late effects.
Distant Follow-up Model	This MOC is where CCS are followed only by telephone, mail or e-mail.
Minimal Follow-up Model	When CCS are not receiving regular follow-up for their cancer.

Transition from Pediatric Care to Adult Care

The health risks faced by CCS continue into adulthood. As such, the transition between pediatric and adult care is likely to occur at most centers. Throughout life, individuals experience an assortment of transitions (29). Blum et al. describe the healthcare transition as the “purposeful, planned movement of adolescent and young adults with chronic physical and medical conditions from child-centered to adult-oriented healthcare systems (30).” Schumacher et al. break down transition into four types: (1) developmental transition of individuals and families (i.e., entering into adult care from pediatric care); (2) situational transition (i.e., moving out of parents house and into independent residence); (3) health and/or illness transition (i.e., diagnosis of a health condition, or completion of a treatment or therapy); and (4) organizational transition (i.e., implementation of new policy or practice or change in leadership) (31). Transition of a patient can include one or more of the above-mentioned types of transitions at any given time (31). Any definition of transition highlights an event, a process, or a change over time. Transition generally results in moving from a situation of comfort to a level of unfamiliarity in adulthood, leading to a restructuring of an individual’s reality (32).

Transition is a complex and multifaceted process. For adolescents, it is further complicated and influenced by changes in physical and emotional development (31, 33). Adolescents are at a critical point in their lives where many of the health behaviors that will last their lifetime are being formed (30, 34, 35). Research suggests that healthcare transitions do not occur in isolation, but rather are entrenched within developmentally appropriate and individualistic progressions (36). In addition to laying a foundation for future health patterns, this time in a person’s life is one of vulnerability because of all the layers of change individuals are faced with. At no other time than birth will an individuals' experience so much change as they do when moving from adolescence to adulthood (35). There are many challenges that come with healthcare transitions. Therefore, it is important that researchers find what works best in their respective fields and how to maximize interventions and programs that support young adults with chronic conditions during the transition period.

The Issue with Transitions for CCS

Like all specialized medical needs, childhood cancer survivorship raises unique issues related to transition. CCS are unique, in that despite surviving their disease, they continue to battle health risks. Many CCS want to put their cancer behind them and look forward to moving into a new normal after cancer. CCS were still developing when they received their treatments and the detrimental effects of these treatments often will not be observed until later in life (7, 8, 37). Transition is further complicated by the difficult timing of CCS moving out of adolescence and into adulthood and the complex nature of this time in any person's life (20).

An optimal transition is achieved when CCS are prepared to move into adulthood and receive continued medically and developmentally appropriate care (38). Adequate transition should address the physical and mental health of CCS, as well as their developmental, educational, vocational, social and financial needs (39). As CCS prepare for transition, they should have knowledge of their cancer, medications and other treatments, and potential long-term risks (39). They should have developed the skills to manage their own care moving forward into adulthood.

Many barriers pertaining to transitions are outlined in the literature. According to Hergenroeder et al. there are four key barriers in care: patients not being prepared or ready to transfer; pediatric providers not prepared to participate in proper transition processes; adult systems not adequately prepared to meet the transitioning issues of adolescents with special needs; and insufficient communication between primary and specialist care (34). The complex nature of transitions makes it challenging to understand the barriers in their entirety because there are so many layers. Further complicating the matter is that CCS are coming from a family-centric system where decisions are made as a team and more comprehensive health services are provided. It can be a challenge to enter adult healthcare where autonomy is a necessity (See *Table 2.2* for details of the differences from pediatric and adult care) (40, 41). CCS will all have different experiences within health systems. It should be noted that these differences between pediatric and adult care are general differences that have been shared by adolescents (40).

Table 2.2 *Differences between Pediatric and Adult Healthcare System*

Pediatric Healthcare	**Adult Healthcare**
Family focused	Individual focused
Many resources provided and arranged through the pediatric system (i.e., social work, psychologist)	Individual is responsible for arranging additional services and appointments
Doctor will talk to parents/caregiver about their care	Doctor talks directly to individual and expects them to make decisions about their own care
Healthcare team spends time getting to know patient and their family	Can be challenging to develop a relationship with healthcare providers
Parent involvement in care is expected	Adults over the age of 18 years must grant permission (if desired) for parents to be involved in care
Parents advocate for patient	Individuals advocate for themselves

Without a proper transition, CCS miss out on developing relationships with their adult care team as well as recommended screening practices. Absence of these high-quality transitions leads to a more reactive rather than proactive treatment of health issues (2, 42). Overall, across key stakeholders, there is a lack of awareness of the need for transition planning which leads to CCS missing out on aftercare in adulthood (42). In fact, it is estimated that more than half of adolescents with a chronic condition report that services are inadequate during their transition into adult care (43). CCS are missing out on aftercare because of this misstep. While transition is a fundamental component of continued aftercare, it should be noted that other factors may also play a role in quality aftercare.

The need to ensure continuity of care through transition is widely acknowledged. Despite this, there is a paucity of literature that highlights best practices. From a system level, policies and guidelines should be in place that supports transition (39).

These policies should be formed from evidence-based research that support ongoing education and training for all key stakeholders. General guidelines exist for transition practices; however, they are not childhood cancer specific and do not take into consideration the specific issues faced by the population at hand. These guidelines will be discussed later in this Chapter. With proper initiatives, such as CCS specific guidelines, in place to support CCS in their transition, we can potentially mitigate health risks later in life.

Transition Guidelines for Children with Chronic Conditions

In Canada, children with chronic illness are getting lost in the system when they leave pediatric care (44). Key stakeholders in Canada, including the Canadian Pediatric Society (CPS) and the Society of Adolescent Health and Medicine, have recognized this deficit and provide recommendations for transition programs (44, 45). Further, they discuss developmentally appropriate transition processes as an integral part of the pediatric system. Coordination between HCP and families is paramount for the success of this preparation. Key stakeholders in the US, including the American Academy of Pediatrics (AAP), the American Academy of Family Physicians and the American College of Physician-American Society of Internal Medicine, also place emphasis on the value of transitions (46). Other examples exist globally, such as a consensus statement on successful transition from pediatric to adult care for adolescents with chronic conditions presented by the Adolescent Health and Medicine Working Group of the European Academy of Paediatrics and a report on the Key

Principals of Care for Young People Transitioning to Adult Services by the Centre for Adolescent Health , Royal Children's Hospital (47, 48).

Position statements provided by the CPS and the AAP outline several principles for a successful transition (46, 49). These include adolescent involvement in their healthcare needs; adolescent and family understanding of their healthcare needs; understanding of personal potential for education, vocation, recreation, and activity; completion of adolescent developmental tasks; and the attainment of self-esteem and self-confidence (49). The CPS has also developed general recommendations, including involving pediatricians during the transition process and providing appropriate resources (49). Families and individuals need to take ownership of transition planning and provide appropriate support throughout the process. Moreover, they suggest that transition should be integrated into a clinical setting and be step-wise. Adolescents should be given all relevant information in relation to their diagnosis and treatment as well as skills training for navigating the healthcare system (41, 49). General transition guidelines provide broad standards for children with a specialized healthcare need. The CPS goes as far as to provide a framework for transition and tools for all stakeholders in order to stress the importance of adequate transition (45). The AAP have stressed a "vision of a family- centered, continuous, comprehensive, coordinated, compassionate, and culturally competent healthcare system that is as developmentally appropriate as it is technically sophisticated (46)." Each chronic condition has its own particularities that need to be addressed. The next section will focus on guidelines specific to CCS.

Guidelines for Childhood Cancer Survivors

There is a need for CCS-specific guidelines to address the specific issues faced by CCS. The Children's Oncology Group (COG), a clinical trials group supported by the National Cancer Institute, has put forth the *Long-Term Follow-Up Guidelines for Survivors of Childhood, Adolescent, and Young Adult Cancers* (18). These guidelines have been developed and published by the COG Late Effects Committee, nurses, and the patient advocacy committee using an evidence and consensus-based process (15). The goals of the COG focus on promoting healthy lifestyles, continuing long-term health follow up, taking part in proper screening and surveillance of late effects and promotion of interventions for late effects. This is a guide for healthcare providers who are delivering care to CCS in a clinical setting (18). This resource also offers patient education material to help enrich follow-up visits and understand Long Term Follow-up (LTFU). Unfortunately, there are limited resources specifically dedicated to helping clinicians ensure that CCS do in fact transition in LTFU. The Pediatric Oncology Group of Ontario (POGO) offers some guidance online for CCS. The POGO childhood cancer resource page lists a number of programs and services including aftercare clinics and transition support for CCS (50).

The Canadian Cancer Society also provides a detailed and informative website for all areas of cancer, including aftercare for CCS (51). They stress the importance of aftercare and managing late effects; however, there are no guidelines for transitioning CCS into adult care. The Canadian Cancer Society does discuss different types of transitions and provide more reading material, a toll-free number and a form to request more information.

Interventions for Aftercare and Transitions

According to the WHO, a health intervention is “an act performed for, with or on behalf of a person or population whose purpose is to assess, improve, maintain, promote or modify health, functioning or health conditions (54)”. Some institutions have dedicated time and resources aimed at promoting interventions for aftercare. These interventions are usually conducted in an effort to improve population health on some level. This includes issues related to health promotion, psychosocial needs, transition, education, physical activity (PA) and other health behavior modification interventions (8, 41, 49, 55). Generally, interventions found in the literature are often focused on addressing gaps in the care of CCS. In the studies that we have reviewed, interventions are generally offered once in an effort to support CCS at a vulnerable time in their lives. Some examples of these interventions include mobile applications, educational forums and booklets (56-68).

Interventions can be supportive for patients and their families. When individuals have a sense of self-efficacy and autonomy and believe that they can complete a task, they are most likely to stay committed and follow through (69, 70).

Interventions to improve the transition from pediatric to adult care for CCS are considered important; however, the number of interventions is still limited and evidence demonstrating their effectiveness is lacking. Despite improved health outcomes being linked to transitioning practices, there are limited interventions that support CCS at this critical time (39, 41, 46, 49). More research needs to be carried out in this field to better understand the role of interventions to support the transition into adult care for CCS. To date, there are few studies that target interventions at the transition phase for CCS. Chapter 3 highlights the results of a systematic review of interventions to improve the aftercare of CCS. In that chapter, we will discuss in detail examples of interventions and their evaluations.

Framework for this Research

There is an opportunity to improve substantially on the quality of our healthcare system, particularly when it comes to transitions. In this section, I present the framework used to guide the selection and development of our intervention. A number of approaches are identified in the literature to guide care improvement processes, including the *Triple Aim Framework* and Donabedian's *Quality of Care Framework* (71, 72). Ultimately, we chose the WHO's *Quality of Care: A Process for making Strategic Choices in Health Systems* framework because it presents a structured process for improving quality of care within the healthcare system and offered a step-wise comprehensive approach for identifying a context-appropriate, evidence-based intervention for improving care for a specific population (73).

The WHO framework is a capacity-building tool that focuses on developing systematic processes that lead to improved outcomes for specific populations (73). This framework is previously established for developing interventions in healthcare. The WHO strategy aims to provide stakeholders with an opportunity to make informed strategic choices which also incorporates the available evidence on a topic (73). The framework aligned with our need to complete a comprehensive study and clearly states that accountability for quality in healthcare can be dispersed in a variety of ways.

The WHO framework has three fundamental steps: analysis, strategy and implementation (Figure 2.1). Analysis requires key stakeholders to be involved and a situational analysis of the current system to be conducted. The analysis process should be comprehensive and include current systems, policies and priorities. For this dissertation, as part of our analysis of the issue, we completed two systematic reviews and a case study. The information acquired help determine the specific needs of the population. The second step, strategy, focused on determining an intervention that can satisfy the domains of care. This step was completed using the information gleaned from our qualitative interviews alongside the WHO domains. Finally, the implementation step, involved delivering the chosen intervention. An evaluation of our intervention was important to ensure it met the needs of the population. The intervention used previously gleaned evidence to support CCS in NL transitioning from the Janeway to adult care.

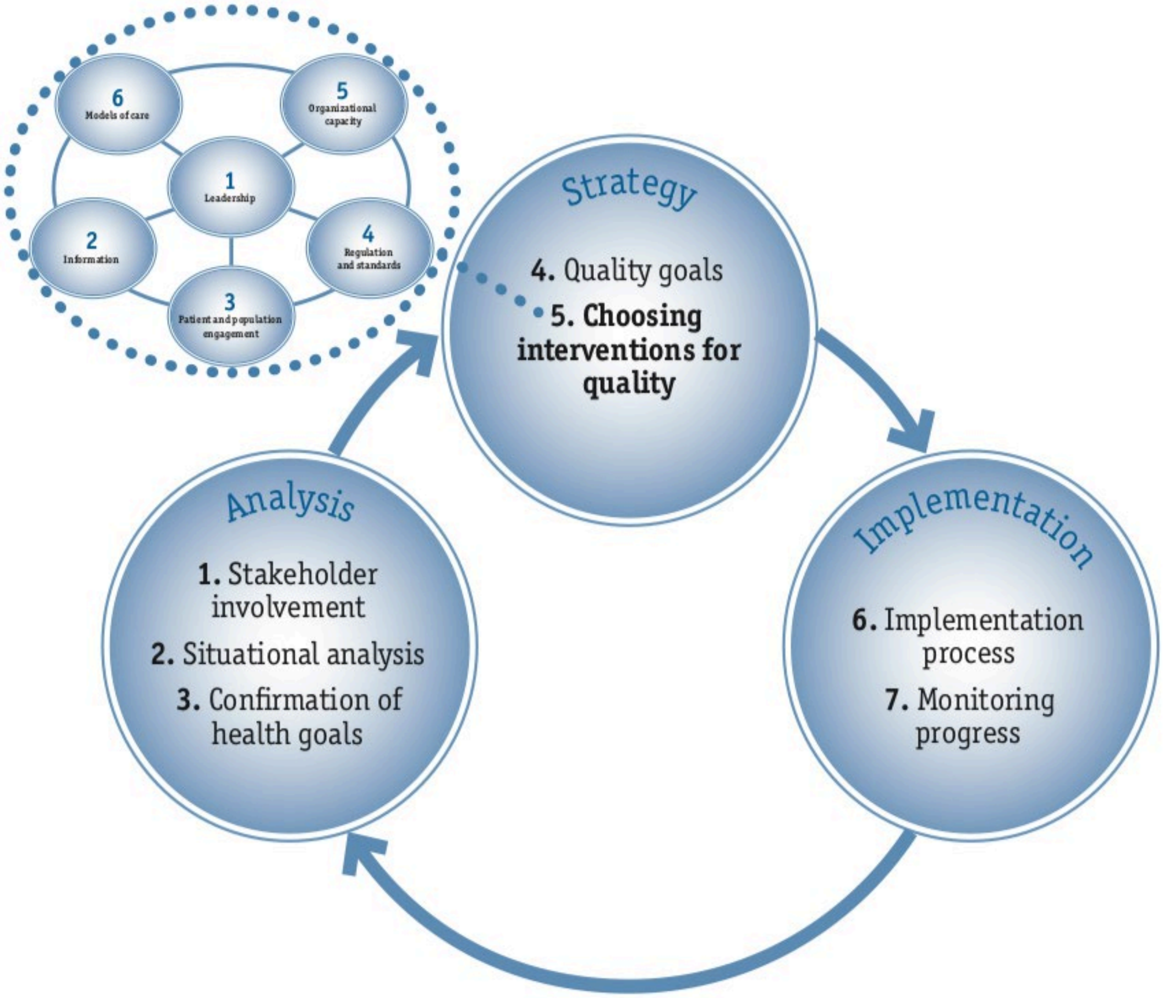

Figure 2.1 *Linking Domains with the Decision-Making Process*(73).*

*Permission to use image provided by the WHO on November 25, 2019

Closing Remarks

The aim of this project is to understand and improve the transition into aftercare for CCS in NL. Advances in the treatment of childhood cancer have led to high survival rates for this population. In turn, more CCS will be needing aftercare in adulthood to manage the potentially threatening late effects of their cancer treatment. Despite the knowledge surrounding these future health risks, many CCS are not receiving adequate care. In this project, an accurate depiction of how transition of CCS is occurring in NL to improve service integration is described, to provide recommendations and to develop supports that may be implemented within the current healthcare environment to improve the organization and quality of care for cancer patients.

Minimal empirical evidence exists related to transitions and CCS; therefore, addressing this area is the primary focus of this dissertation. The transition from pediatric to adult-centered care has become an important issue and one that aligns closely with addressing the concerns related to aftercare for CCS. In many ways, this project could have a significant impact on the care of this population. Given that similar issues of aftercare and transition are faced by other pediatric populations who have chronic conditions, it is hoped that the lessons learned through the project may be shared by other pediatric programs.

The remaining chapters of this dissertation will present completed research, including two systematic reviews and categorization manuscript, a case study and the development and evaluation of our intervention. This research coupled with the gaps identified within the literature support the need to enhance the coordination, continuity and provision of healthcare services that promote optimal health functioning and quality of life (QOL) for this population. When transition occurs in a coordinated and continuous manner, transitioning does not have to be a health crisis but rather a developmental milestone.

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Chapter 3

Childhood Cancer Survivorship: Categorization, Models of Care and Interventions

Introduction and Overview

As outlined in Chapter 1, I chose to use the *WHO's Quality of Care: A process for making strategic choices in health systems* as a guiding framework for this research. As such, the first step was to complete a situational analysis, which includes a thorough review of the relevant academic literature. The initial plan was to conduct a systematic review around the evidence supporting different models of aftercare. However, there was an issue related to the fact that models of aftercare were categorized in various ways within the current academic culture. The first article of this chapter is a published commentary which argues for standardizing the categorizations of MOC for CCS. This article helps to better interpret various MOC in the field and maximize the applicability of the available evidence. The second article is a systematic review of MOC for CCS. This review compiled the literature on various MOC and helped to describe the continuum of care in the field and allowed me to build a context for CCS in NL in the following chapter. The third article is a systematic review that summarizes the evidence for the effectiveness of interventions for improving the transition and/or aftercare for CCS. This review helped inform the research when choosing an appropriate intervention to improve care for the NL population.

The manuscripts included in the chapter are the published or drafted for publication versions of these articles. Because of journal restrictions on article length, some material could not be included. To address this issue, I included additional methodological information by each article in *Appendix A*.

Co-authorship Statements

Standardizing the Categorizations of Models of Aftercare for Survivors of Childhood Cancer

- Devonne Ryan designed the research, supervised by Dr. Roger Chafe and Dr. Paul Moorehead.
- Devonne Ryan wrote and drafted the manuscript, supervised by Dr. Roger Chafe.
- Devonne Ryan critically appraised the drafted manuscript, supervised by Dr. Roger Chafe and Dr. Paul Moorehead,.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead all read and approved the final manuscript.

Models of care for childhood cancer survivors once they become adults: A Systematic Review

- Devonne Ryan designed the study supervised by Dr. Roger Chafe and Dr. Paul Moorehead.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead worked together to select studies.
- Devonne Ryan drafted the manuscript, supervised by Dr. Roger Chafe.
- Devonne Ryan, Drs. Roger Chafe, Paul Moorehead, Kevin Chan, Katherine Stringer, and Kathleen Hodgkinson were involved in critically appraising the drafted manuscript.

- Devonne Ryan, Drs. Roger Chafe, Paul Moorehead, Kevin Chan, Katherine Stringer, and Kathleen Hodgkinson read and approved the final manuscript.

Interventions to Improve the Aftercare of Survivors of Childhood Cancer: A Systematic Review

- Devonne Ryan designed the study supervised by Dr. Roger Chafe and Dr. Paul Moorehead.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead worked together to select studies.
- Devonne Ryan drafted the manuscript supervised by Dr. Paul Moorehead
- Devonne Ryan, Drs. Roger Chafe, Paul Moorehead, Kevin Chan, Katherine Stringer, and Kathleen Hodgkinson were involved in critically appraising the drafted manuscript.
- Devonne Ryan, Drs. Roger Chafe, Paul Moorehead, Kevin Chan, Katherine Stringer, and Kathleen Hodgkinson read and approved the final manuscript.

Standardizing the Categorizations of Models of Aftercare for Survivors of Childhood Cancer

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Abstract

Background: With significant improvements in the survival rates for most childhood cancers, there is increased pressure to determine how follow-up or aftercare for survivors is best structured.

Main Body: Previous work in this area has not been consistent in how it categorizes models of aftercare, which risks confusion between studies and evaluations of different models. The adoption of a standardized method for classifying and describing different models of aftercare is necessary to maximize the applicability of the available evidence. We identify some of the different ways models of aftercare have been classified in previous research. We then propose a revised taxonomy which allows for a more consistent classification and description of these models. The proposed model bases the classification of models of aftercare on who is the lead provider, and then collects data on five other key features: which other providers are involved in providing aftercare, where care is provided, how are survivors engaged, which services are provided and who receives aftercare.

Conclusion: There is a suitable level of interest in the effectiveness of different models of aftercare. Future research in this area would be assisted by the adoption of a shared taxonomy that will allow programs to be identified by their structural type.

Background

With the significant rise in the number of survivors of childhood cancer, increased attention is being given to how to structure the aftercare for these patients who face lifelong health risks (1-3). Following the treatment of their active cancer, aftercare is initially provided by their pediatric oncology care team. In early adulthood, many survivors transition out of pediatric care, with subsequent aftercare being structured in significantly different ways across cancer programs (4). One focus of research in this area has been on the effectiveness of different models of post-transition aftercare in supporting and serving survivors of childhood cancer (5, 6). Yet in studying these different models of aftercare, conflicting categorizations have been employed, often without much consideration seeming to be given for the adoption of different basis for distinguishing models of aftercare. In this article, we will review the different ways models of aftercare have been categorized across various studies, and then propose a new categorization which allows for a more specific and standardized identification of program types. Finally, we discuss the possibility of employing a similar method of categorization for studying models of post-pediatric care for other chronic conditions.

Main Body

Previous Categorizations

A MOC describes the structure and type of services provided to patients with a particular condition during a period of time or phase of their disease. It broadly "defines the way health services are delivered" for a group of patients (7, 8). Given the multiple factors that are incorporated in any model of care, it is clear that different aspects can be used to distinguish models from each other. In studying the health services available for survivors of childhood cancer, authors have previously used various aspects to distinguish models of aftercare (*Table 3.1*). One approach identifies models that are commonly employed. For example, the Institute of Medicine's *From Cancer Patient to Cancer Survivor: Lost in Transition* took a comprehensive look at healthcare issues for both survivors of adult and childhood cancers (8). In discussing models of aftercare, the authors of the report focus on "promising models of follow-up care," and examines a shared-care model, a nurse-led model and survivorship follow-up clinics. Similarly, in their survey of pediatric oncology centres, Eshelman-Kent et al. employ a list of models "identified in the literature (4)." Specifically, they propose using the following categories: Cancer Center-Based Model Without Community Referral, Community Referral Model, Hybrid model (Combined Cancer Center and Community Based Model), Postal/Internet/phone-based model, Adult oncologist and Other. In their Delphi survey of policy experts, Mertens et al. used the categories chronic disease model, primary care model and late effects model (9).

An alternative approach has been to identify models of care in terms of the setting in which the care is given. Oeffinger and McCabe have previously taken this approach, evaluating models of care in terms of whether they are hospital-based, community-based, or shared care, i.e., include both community and hospital care (6). In examining the use of care plans for both survivors of childhood and adult cancer, Hahn and Ganz also distinguish models of aftercare in terms of setting: an academic medical center, a community hospital, a primary-care medical group and a county hospital (10). Wallace et al. and Michel et al. distinguish models in terms of the profession of the person who is the lead for organizing care, e.g., medically supervised late effects clinic, primary care physician-led or nurse-led, and postal or telephone follow-up, with the appropriate level of care being dependent on the risks associated with the survivor's type of cancer and treatment received (2, 11). Heir et al. use a blend of different aspects to distinguish programs, including in terms of communication modalities (e.g., face-to-face clinic visits, telephone, postal, email or SMS/text-based); physician versus nurse-led follow-up; and hospital versus PCP follow-up (5).

In their 2003 Institute of Medicine report, Weiner, Simone, and Hewitt offer another approach to categorizing a MOC (12). They identify the "comprehensive survivorship program" model. Comprehensive programs are those that have "a dedicated time and place for the clinic, met at least twice a month, were staffed by a doctor with experience in the late effects after treatment for childhood cancer, had a nurse coordinator, provided state-of-the-art screening for individual's risk of late effects, provided referrals to appropriate specialists, and provided wellness education." Hewitt, Weiner and Simone also discuss this type of program (12). Similarly, Aziz et al. has found this MOC is commonly employed by larger pediatric oncology programs in North America (13).

Table 3.1 *Previous Categorizations of Models of Care for Survivors of Childhood Cancer*

Author	**Basis of Categorization Identified by the Author**	**Models of Care Identified**
Hewitt, Greenfield, Stovall(8)	"promising models of follow-up care,"	-Shared-care model -Nurse-led model -Survivorship follow-up clinics
Eshelman-Kent et al.(4)	"models identified in the literature"	-Cancer center-based model without community referral -Community referral model, hybrid model (combined cancer center and community-based model) -Postal/internet/phone-based model -Adult oncologist
Mertens et al.(9)	Not identified	-Chronic disease model -Primary care model -Late effects model
Oeffinger and McCabe(6)	Setting of Care	-Hospital-based model -Community-based model -Shared care model
Hahn and Ganz(10)	Setting of Care	-Academic medical center -Community hospital model -Primary-care medical group -County hospital
Wallace et al. / Michel et al.(2, 11)	Lead Provider	-Medically supervised late effects clinic -Primary care physician-led model -Nurse-led model -Postal or telephone follow-up model
Heir et al.(5)	Communication modality / Lead Provider / Setting	-Face-to-face clinic visits -Telephone, postal, email or SMS/text-based model -Physician versus nurse-led follow-up -Hospital versus primary care follow-up
Hewitt, Weiner and Simone (12)	Identified in the literature	-Comprehensive survivorship program
Aziz et al.(13)	Identified in the literature	-Comprehensive survivorship program

There is nothing inherently wrong in adopting either of these approaches for distinguishing models of aftercare. But given that there is insufficient evidence around which models are the most appropriate, (2, 5) the lack of a clear and consistent method of categorizing models of aftercare risks defusing the evidence that is available. A standardized method of categorization would allow for more accurate description of programs. It would make explicit the defining aspect of similar categorizations used in previous studies, e.g. primary care physician-led, primary care follow-up, community-based and community referral model--which are all likely referring to the same or very similar types of programs. In order for progress to be made in the evaluation of models of aftercare, there needs to be a standardized way for classifying and describing various models of aftercare for survivors of childhood cancer, particularly across studies.

A New Taxonomy

There are relatively few examples of classifications of models of care having been developed even in other disease areas. Those that have been reported on in the academic literature, e.g., relating to maternal care or community-based mental health services, have used multi-year approaches to engage a range of stakeholders on how models should be defined and to define the data elements to capture in administrative systems (14, 15). These studies were done in the context of reporting data to government agencies related to evaluations of outcomes and payments for services. We developed our proposed taxonomy in the context of planning a systematic review of models of aftercare. In developing our classification, we ran into the same issue as faced by others. In particular, the need to balance the development of a system of classification that can identify features that can meaningfully group programs together, while capturing the “level of granularity” about programs required to conduct an appropriate evaluation between them (14).

In developing our taxonomy, we first reviewed previous categorizations and identified key program features they included. Based on this review, we identified six fundamental features: 1) the provider primarily responsible for managing aftercare; 2) the other providers who are regularly involved in providing aftercare; 3) the location of care; 4) the method of engaging survivors, including how survivors receive aftercare and how a program tracks its survivorship population; 5) the aftercare services provided; and 6) who receives care through the aftercare program, e.g., whether a program is risk stratified or focused only on a select group of survivors. There are clearly other relevant program characteristics that impact the care survivors of childhood cancer receive, including age restrictions on follow-up; frequency of follow-up; available resources for the program; whether transition occurs within the same institution, e.g., between the pediatric and adult oncology programs within the same cancer hospital, or to a different institution; and whether research and evaluation are part of the program. In not including these features in our framework, we do not mean to imply that they are not important or that they cannot also be captured depending on the study aim. Rather we hope to develop a framework for categorizing models of aftercare that can provide a consistent way of characterizing different types of aftercare programs without being too restrictive. For example, one of our concerns with the definition of comprehensive survivorship programs is that it is too detailed in its criteria to include anything other than these specific programs (12).

The first feature of our categorizations is the specialization of the provider who is primarily responsible for providing and organizing aftercare. Aftercare is initially provided by the pediatric team, but after adolescence cancer programs differ in terms of who is responsible for providing care. Eshelman-Kent et al. report that 35% of survivors continue to be followed into adulthood by the pediatric care team (4). If survivors are followed by a pediatrician or a pediatric long-term care clinic into adulthood, we would classify this model as a pediatric-led model of aftercare. For models in which the survivor transfers to a new care team, it may be an adult oncologist, a PCP, or a nurse who is the provider primarily responsible for overseeing the survivor's aftercare. Another common arrangement is a hybrid where a PCP follows the survivor for their survivorship care, but this physician maintains a close connection to an oncology program that can be called upon if any serious issues arise (1). For models of care which follow survivors only by telephone, mail or e-mail, we would classify these models as distant follow-up. If there is no regular follow-up with survivors, we would classify these models as minimal follow-up.

Table 3.2 shows our proposed categorization. There are a number of reasons for starting with the provider as the primary basis for classifying different models of aftercare. First, it is a basis for distinguishing models of aftercare used, or partly used, by other authors (2, 5, 11). It also incorporates the main basis used for distinguishing models of aftercare. For example, those who classify models of aftercare in terms of the setting where the care is given are likely really concerned with whether the survivor is receiving follow-up specialist care versus primary care. Furthermore, identifying the provider makes it more specific the level of specialization the provider has, e.g., whether they are a nurse, a pediatric oncologist or adult oncologist, if survivors are being followed in the hospital setting. In most situations, it should not be too difficult to determine who is the main provider responsible for aftercare within a particular program. Even for those survivors seen in a survivorship clinic, researchers and program directors should be able to identify the person who is ultimately responsible for coordinating care in most cases.

While identifying the lead provider as the basis for classifying models of care, as stated above, there are other relevant features that are useful to include in identifying models of aftercare. Yet, rather than developing specific categories for each of these features, in practice we found that this approach led to many difficult choices in categorizing programs. Instead of forcing these artificial distinctions on programs, and thereby missing much of the relevant information, for the other five features we simply capture the detail information around each. Our proposed approach then is to classify programs broadly in terms of their lead provider, then identifying other key aspects where details need to be captured. It is hoped that this approach will allow for some groupings of relevant programs, while allowing for the appropriate level of detail to be able to distinguish key program features.

Table 3.2 *Proposed Categories of Models of Aftercare*

			Models of Care
Models of Aftercare	1. Which provider is primarily responsible for aftercare?	a) Pediatrician	a) Pediatric-Led Model
		b) Adult Oncologist	b) Adult Oncology-Led Model
		c) PCP	c) PCP-Led Model
		d) PCP and Oncologist	d) Hybrid Oncology/PCP Model
		e) Nurse	e) Nurse-Led Model
		f) Phone/Text/E-mail	f) Distant Follow-up Model
		g) None	g) Minimal Follow-up Model
Other Key Features	2. Which providers are regularly involved in providing aftercare?		
	3. The location of care		
	4. How are survivors engaged?		
	5. Which services are provided?		
	6. Who receives services?		

Discussion

In developing our approach to categorizing the different models of aftercare, we have tried to incorporate features that other authors have seen as important. For this reason, it should be able to be applied to categorize programs reviewed in previous studies even if they used a different basis of categorization. We have also tried to format the approach using questions that can be clearly answered, so that this approach should be fairly straight forward to use. While it is possible that questions may arise whether a program is multidiscipline or who is the lead provider, in practice program characteristics around these key categories should be identifiable for most programs.

Still, given the variation in programs that are possible, it is likely that questions of proper categorization will arise. For example, if you had an aftercare program in a pediatric hospital the lead physician of which is a primary care physician by training, under our classification this would be PCP led, even though the provider would have a good deal of specialty knowledge. If the lead physician were to change, this may result in a reclassification of the entire model. Those reporting on different models of care should be cognisant of these factors and report on them when they do occur.

We developed this categorization based on the articles we identified in a systematic review on models of aftercare for survivors of childhood cancer. It is possible that we have not identified in this review all the potential models of aftercare that have been proposed in the academic literature. We also recognize that the Distant Follow-up Model could be seen as a mode of communicating with patients, rather than a type of provider. We have included this model as a type of provider because it is different than regularly meeting with a nurse or other provider, even though there is likely a HCP who is reviewing these responses. The basis for the categorizations we propose seems most reasonable to us given the work that has been previously published on models of aftercare and the way that others have categorized them. Our goal is to highlight the need for a consistent categorization in this area and to propose an approach for doing this. We recognize that other approaches are possible and would welcome further discussion around ways to improve our proposed categorization and to maximize the utility of the research being conducted on this topic.

Survivors of childhood cancer face a number of unique health risks that make their required aftercare unique from other patient groups, including adult cancer survivors. We know that poorer health outcomes are associated with unmet needs of CCS and families during the period when they need aftercare (3). Optimal participation and structure of models of aftercare offers CCS an opportunity to enhance their healthcare and ultimately reduce their risks of late effects associated with their cancer treatment. Yet issues around transition and follow-up care occur for many groups of patients, including most pediatric patients with chronic conditions who require ongoing follow-up. The model that we propose here for categorizing models of care for survivors of childhood cancer could be modified to be applied to other patient groups. For example, for patients with type 1 diabetes similar issues arise regarding the specialization of the provider who is responsible for managing their condition, and around continued access to multidisciplinary care. How structured programs are and whether they stratify patients based on their level of risk or acuity are also questions faced in other areas. Research in these areas should explore the possible adoption of a method of categorization similar to the approach we are proposing for survivors of childhood cancer.

Conclusions

There is a sizable amount of interest in the effectiveness of different models of aftercare. Future research in this area would be assisted by the adoption of a shared taxonomy that will allow programs to be identified by their structural type (5). Because of existing staff, resources, and geographic location, there may be little flexibility regarding the adoption of new models of aftercare. Regardless, programs could likely implement interventions which have been evaluated in programs with similar structures. Reviews, like Singer et. al, on specific models maybe a step in this direction (16). Yet at the very least the adoption of a standardized method for classifying different models of aftercare will help avoid unnecessary confusion and help ensure the maximum utility of the evaluations that are conducted.

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Models of care for childhood cancer survivors once they become adults: A Systematic Review

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This version of the article has been drafted for publication.

Abstract

Background: After surviving cancer, CCS are at increased risk of morbidities and mortality from their cancer and treatments. There is a growing population of CCS that require lifelong medical surveillance. Our objective was to examine the evidence supporting clinical impacts, patient/provider satisfaction or program outcomes in different MOC for CCS.

Methods: A review was conducted using PubMed, Embase, CINAHL, ERIC, Cochrane Database and PsychoINFO databases. Studies were included if they (1) described or evaluated health services and programs of care provided to adult CCS; (2) presented original empirical findings; (3) were published between January 1, 1995 and September 13, 2017; and (4) were full articles, published in English.

Results: We reviewed 9400 titles and 440 abstracts, with 22 articles meeting the inclusion criteria. The MOC identified were: pediatric-led, adult oncology-led, PCP-led, hybrid oncology/PCP, nurse-led and other-led follow-up. We found evidence of patient and provider satisfaction, supporting pediatric-led and hybrid oncology/primary care models.

Conclusions: Additional research is needed, including comparative evaluations of different MOC to determine the most effective structures for providing aftercare.

Implications for Cancer Survivors: We found a relatively weak evidence-base in support of a specific MOC. However, authors and study participants considered a MOC with risk stratification, multidisciplinary care, and adequate transition processes to be most suitable.

Background

The last 40 years have seen dramatic improvements in survival rates for patients who have cancer in childhood (1, 2). While the resulting growth in survivors is extremely positive, it is estimated that 2/3 of CCS will exhibit late effects from their cancers and subsequent treatment (3, 4). Late effects include increased risks of developing subsequent cancers, physical morbidities, neurocognitive impairments, psychosocial issues, and fertility concerns (4-9). Given the range of health risks, it is important that survivors receive appropriate follow-up, or aftercare, throughout their lives (10, 11).

Aftercare is initially provided by their pediatric oncology care team; however, when CCS reach late adolescence, aftercare is structured differently across cancer programs (12). Our review updates and expands on previous reviews that were either more limited in scope (13-15) or looked only at specific MOC (16), as well as reviewing new empirical evidence. Our review is also novel in the way that we categorize the MOC (17). Our objective was to examine the evidence supporting clinical impacts, patient/provider satisfaction or program outcomes in different MOC for CCS.

Material and Methods

A systematic review of the literature was conducted following the recommendations of the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analysis) statement (18).

Search Strategy

We performed a structured literature review of PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo between June 26, 2015 and September 13, 2017. The full review protocol is available in the *Appendix A*. The outcomes examined included clinical impacts, patient/provider satisfaction or program outcomes.

Eligibility Criteria

Studies were included if they (1) described or evaluated health services and programs of care provided to CCS; (2) presented original empirical research; (3) were published between January 1, 1995 and September 13, 2017; and (4) were full articles published in English.

Study Selection

All titles were reviewed by two researchers to remove obviously irrelevant titles. The full articles for all included abstracts were read to determine if articles met the study criteria. The reference lists of all included articles were also reviewed to identify if articles were relevant to this review. Articles were excluded primarily because they did not include empirical research or failed to describe or evaluate health services or programs of care provided for CCS.

Data Extraction

A data abstraction form was developed and included a description of the study, study type, MOC /program, geographic location, number of participants, type of study participants, key features of MOC studied, outcomes evaluated, and main conclusions (*Table 3.3*).

Quality Assessment

We assessed the methodological quality of all included articles using critical appraisal guidelines developed by Strengthening the Reporting of Observational studies in Epidemiology (STROBE) (18). Articles were not excluded because of quality issues, but study limitations are discussed in the Results section.

Results

Search Results

Our search initially identified 9400 articles and 8960 were excluded on initial review of titles. Abstracts were reviewed for 440 articles, which further excluded 382 articles. Full text review and reference searches were completed on 58 articles. Of these articles, 36 were rejected, leaving 20 articles. Articles were excluded because they did not provide empirical research or did not examine the outlined outcomes. Two additional articles were identified by reviewing the references of the full-text review articles (*Figure 3.1*).

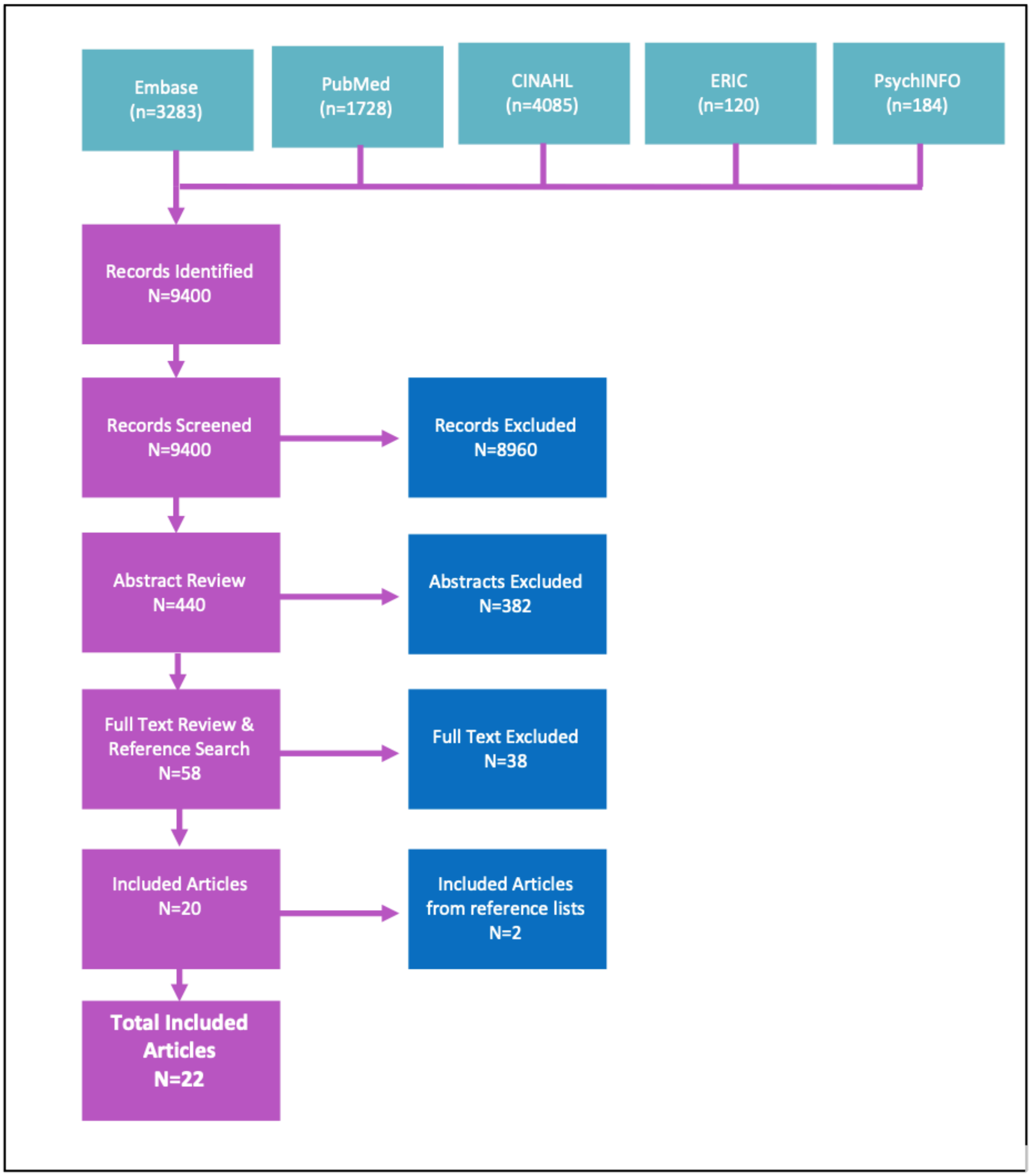

Figure 3.1 *Search Strategy for MOC for CCS*

Study Characteristics

Most articles that met the inclusion criteria (n=18; 82%) were published after 2005. Studies were most often conducted in the USA (n=7; 32%), followed by studies in both USA and Canada (n=4; 18%), the UK (n=4; 18%), Switzerland (n=2; 9%), Netherlands (n=2; 9%), Norway (n=1; 5%), Australia and New Zealand (n=1; 5%) and Canada (n=1; 5%). The type of study participants varied and are illustrated in *Table 3.3.* The programs identified in this review are described using the categorization model outlined in the introduction of this chapter. This table can be found in *Appendix B*. Examples of study outcomes found in this review include: patient participation in aftercare, access to aftercare, type of MOC, patient satisfaction, HCP involvement in care, and willingness of patients and/or providers to participant in aftercare practices.

Pediatric-Led Model

In a pediatric-led program, a pediatric oncologist or program is responsible for organizing and providing aftercare. Typically these programs are located in a pediatric hospital (19, 20). Oeffinger et al. surveyed members of the *Children's Cancer Group* and *Pediatric Oncology Group* in the U.S. and Canada. They found that 53% of these programs have some type of aftercare clinic, and that 44% of these clinics provide aftercare to CCS (21). They also found that 93% of programs used a pediatric oncologist (21). Kenny et al. surveyed pediatric oncology programs in the U.S. and found that 11 out of 12 programs had aftercare clinics. All 11 programs were staffed by a pediatric oncologist (22). Sadak et al. found that 25% of survivors in their pediatric-led aftercare program received their treatment at another cancer institution, coming to their program only for aftercare (23). As part of the Swiss Childhood Cancer Survivor Study completed by Vetsch et al., parents of CCS assessed follow-up care. The majority of survivors (79%) received follow-up care from a pediatric oncologist (24). Michel et al. used data from the Childhood Cancer Follow-up study in Switzerland to describe the involvement of Swiss physicians in follow-up care (25). Of the physicians that participated, 85% reported the involvement of a pediatric oncologist in follow-up care (25). Another study by Signorelli et al. used semi-structured telephone interviews with LTFU clinics across Australia and New Zealand. They found that various models were used across all LTFU clinics (26). Majority were led by a pediatric oncologist, with the remainder being nurse-led (26). Finally, Ristovski-Slijepcevic et al. surveyed all pediatric programs in Canada. The authors found that 71% of programs had pediatric-led aftercare programs; however, only 35% had continued access to aftercare once they reached adulthood (20).

The pediatric-led model has high levels of both patient and provider satisfaction. Michel et al. surveyed CCS and found that they favored aftercare with their existing pediatric oncologist, but they recognize this arrangement may not always be feasible (27). Aziz et al. surveyed directors of LTFU programs in Canada and the U.S. and found that 83% were pediatric oncologists. Most program leaders discussed that pediatric-led multidisciplinary MOC could be suitable for LTFU (19).

A number of issues have also been identified with the pediatric-led model. Aziz et al. found that most programs focused only on survivors younger than 25 years. There were issues around insuring aftercare for older adult survivors and that pediatric providers expressed “a lower clinical comfort level” when following adults(19).

Oeffinger et al. found that pediatric-led multidisciplinary LTFU clinics reported patient uncertainty about the need for follow-up, low levels of attendance by survivors, and difficulties retaining a connection with adult survivors (21). The most common concern relates to whether all patients need to be continued to be followed by a multidisciplinary clinic and whether there are sufficient resources for these clinics to provide adequate care to all CCS. Michel et al. suggested that having all survivors attend multidisciplinary pediatric-led clinics maybe preferable, but that this arrangement may not be feasible given the increasing number of survivors (27). The LTFU program directors surveyed by Aziz et al. suggested either transitioning survivors to the community or developing young adult programs directed by PCP (19). Overall, pediatric aftercare, led by a pediatric oncologist was a common model of care. It was less common to see these programs continue into adult years. Aftercare was dependent on patient readiness and often adolescents continued their pediatric-led aftercare into adulthood.

Primary care physician-led (PCP-led) model

Nathan et al. surveyed CCS and found that only 14.6 % received aftercare in a cancer centre, with the majority receiving general medical care (8). In another survey by Nathan et al., 48% of PCPs reported rarely receiving a treatment summary from the pediatric centre which treated the patient (28). The authors also discuss that many physicians were not comfortable treating survivors of certain childhood cancers (28). Szelda et al. found that 55% CCS were ultimately transferred from the cancer survivorship program at the Children's Hospital of Philadelphia to adult-focused follow-up. Of these CCS receiving follow-up care, 50% received their care from PCP (29). Oeffinger et al. reported that PCP provided healthcare most often to CCS (30). Similarly, Aziz et al. found that a large number of PCPs with a wide range of interests and experience were providing some aspect of care for their survivors. LTFU program directors in the Aziz study reported attrition by "loss to follow-up" and difficulties in establishing and maintaining communication with each individual community physician about each survivor (19). Generally, many CCS went to their PCP for aftercare. There were several issues outlined including lack of treatment summary from oncologists and issues with retention.

Hybrid Oncology/Primary Care Model

In the hybrid model, PCPs are responsible for monitoring patients' late effects while maintaining a connection to a cancer physician or program. Both Meacham et al. and Nathan et al. found that PCPs are interested in partnering in models of shared care for CCS, but proper supports need to be in place (28, 31). In fact, Nathan et al. found 85% of PCP respondents preferred to care for survivors in consultation with an oncologist or long-term care follow-up program (28). Blaauwbroek et al. surveyed GPs who participated in a postgraduate course on late effects in pediatric cancer survivors (32). They found that 97% of family physicians were willing to participate in a hybrid/shared-care model. Another study by Blaauwbroek et al., randomized CCS to examine the feasibility of a hybrid/shared care model for LTFU(33). They found that 88% of CCS were satisfied with this MOC (33). Meacham et al. delivered lectures to PCPs and developed a website for PCP that provided relevant information for treating

CCS (31). The website had 471 unique visitors from 30 states in its first 12 months of operation. Lie et al. described common MOC including the shared-care model. The researchers concluded that a shared-care model for LTFU was considered best practice for achieving several unmet needs among lymphoma survivors (34). Patients also found the hybrid model improved personal relations with their PCP and increased their knowledge of future risks (21). Overall, CCS reported high rates of satisfaction with hybrid models of care. Researchers noted that this might be the preferred means of delivering long-term aftercare to CCS.

Other Models of Care

Little evidence was identified supporting other models of aftercare for CCS. Earle et al. reported that survivors in Sheffield (UK) preferred aftercare led by an oncology nurse. Participants preferred this model because they found the level of expertise and opportunities for feedback were preferred over a PCP-led model; however, the authors did not expand on this finding (35). Mertens et al. used a Delphi panel of health policy experts to examine MOC. Their top choice was the “chronic disease model” where a multidisciplinary team provides survivorship care under the direction of an adult oncologist (36).

There were three examples of distant follow-up identified as part of the review. A regional cancer program in Birmingham (UK) surveyed PCP about CCS who were lost to follow-up. In their evaluation of the survey, Parkes et al. found postal follow-up with PCP was effective means to monitor health concerns, inform clinical practice, and enable research for survivors not participating in regular follow-up care (37). Parkes et al. conclude that this type of follow-up allows for surveys that could help direct education campaigns (37). Casillas et al. developed and examined the feasibility of a text messaging MOC for follow-up of CCS (38). Participants approved the development of social networks to enhance LTFU and MOC. Distant follow-up may not be the most ideal means of delivering aftercare; however, studies in this review suggest that at very minimal it is a means to continue engagement with this population.

Transition Programs and Planning

Many of the included studies recognized the importance of a systematic transition from pediatric care to adult care for CCS (19, 20, 22, 23, 25, 39). Unfortunately, for those CCS who do not stay with their pediatric provider, transitioning from pediatric care to adult care can be complex and challenging (21). In fact, Aziz et al. reported that program directors of pediatric LTFU clinics often found it "difficult" to successfully transition survivors to PCPs (19). Additionally, Laar et al. used a questionnaire to evaluate predictors of satisfaction with transition (39). They found that while transitions did not have significant impact on patient satisfaction, patients were more satisfied when their pediatric and adult physician worked together in an adult setting during transition (39). In another study, Kenny et al. surveyed pediatric oncology programs in the U.S. and found that 11 of 12 programs had aftercare clinics. All 11 programs were staffed by a pediatric oncologist and only four programs identified a policy for transitioning CCS to specialized adult program or to a PCP(22). Similarly, Ristovski-Slijepcevic et al., reported that in Canada 76% of pediatric institutions provided a treatment summary or survivorship care plan (20). Despite this, only 2 institutions offered a transition program (20). Sadak et al, reported that transition is treated as an active milestone within the MOC; however, they report no predictive tool being used to assess transition readiness (23).

Risk Stratification

Risk stratification models allow for different levels of aftercare depending on the survivor's type of cancer, treatment experience, and likely benefit from aftercare (11). The motivation for stratification comes from a recognition that patients differ in their risks for late effects depending on the type of cancer they had and the type of treatments they received. Michel et al. found that the level of risk a survivor faces did affect their preference for different models of aftercare (27). They also conclude that "many survivors receive more intense follow-up than necessary given their risk stratification." Wallace et al. identify the need for better evidence around late effects of specific cancers and treatment modalities in order to appropriately risk stratify patients but recognizes the need to do so based on the increasing numbers of survivors. In fact, that many survivors may not need the level of follow-up that they are scheduled to receive (11).

Multidisciplinary Programs

Multidisciplinary aftercare has been described as the preferred means for caring for the complex healthcare needs of adult CCS (21, 36). Within more comprehensive models, this approach usually includes a physician, nurse, an administrator, and allied health support (10, 20, 22, 40). Comprehensive multidisciplinary models have been shown in other areas to be a cost-effective way to improve patient outcomes (41).

Table 3.3 *Study Characteristics*

Author/Year	Models of Care Evaluated / Discussed	Location	Other Features	Type of Study	Study Participants	Conclusions
Aziz et al., 2011	Pediatric-Led; PCP-Led	USA and Canada	Multidisciplinary	Cross sectional survey	26 LTFU program directors	• LTFU clinics have many benefits, but face resource constraints. • The greater adoption of PCP-Led models is needed, but there are concerns about identifying PCP with expertise and patients being lost to follow-up.
Blaauwbroek et al., 2008	Hybrid Model	Netherlands	Multidisciplinary	Randomized prospective cohort	Family doctors and survivors	• 88% of survivors were satisfied with the care provided. • Many family doctors remained attached to the program. • Five patients had second malignant tumour discovered during the program. • PCP felt supported by the connection with the pediatric oncology program, but requested a better flow of information flows.
Earle et al., 2005	PCP-Led; Nurse-led	UK	Multidisciplinary	Focus groups	26 CCS and 33 of parents	• PCP-led clinics were not seen as having sufficient specialty knowledge. • Participants preferred nurse oncology-led aftercare for the level specialist expertise and opportunities for appropriate feedback.
Kenney et al., 2011	Pediatric-Led	USA	Multidisciplinary	Cross sectional survey	12 Pediatric oncology programs	• All programs have a pediatric-led multidisciplinary LTFU clinic. • Four programs reported that they transfer survivors to either a

Author/Year	Models of Care Evaluated / Discussed	Location	Other Features	Type of Study	Study Participants	Conclusions
						specialized adult survivorship program or to a PCP. • Other programs retain all patients. Securing funding for resource intense programs, volume of sub-specialty referral, and participation in research were common challenges.
Laar et al., 2013	Adult Oncology-Led	UK	Adult Oncology / Continued care	Cross sectional	143 Survivors of childhood cancer	• Transition to adult care did not impact significantly upon patient satisfaction. • Shorter waits and knowing why participants were attending the clinic increased satisfaction. • Joint work between adult and pediatric cancer professionals enabled adult survivors of childhood cancer to receive highly satisfactory care in adult services.
Meacham et al., 2012	Hybrid Model; PCP-Led	USA	Multidisciplinary	Interviews; education sessions; implementation and evaluation of online resource.	Physicians	• Participants reported moderate to low familiarity with survivorship issues, but were interested in learning more about supporting CCS. • A website with CCS information aimed at PCP and patient information was developed. • The website had 471 unique visitors in the first 12 months of operation from 30 states. • The authors conclude that PCP involvement in survivor care alleviates some barriers to care such as geographic distance to the cancer center and ensures that more pediatric

Author/Year	Models of Care Evaluated / Discussed	Location	Other Features	Type of Study	Study Participants	Conclusions
						cancer survivors receive recommended coordinated surveillance for late effects of cancer therapy.
Mertens et al., 2004	Hybrid Model; Pediatric- Led; PCP-Led Model	USA	Multidisciplinary	Qualitative, delphi process	Healthcare policy experts	• PCP were unfamiliar with the health problems of survivors. Survivors were often unaware of their risks. • The recommended MOC would incorporate a PCP-Led chronic disease management model.
Michel et al., 2009	Pediatric-Led; Distant follow-up; Nurse-Led; PCP-Led	UK	Multidisciplinary	Pre-and post-visit survey	112 Survivors of pediatric cancer	• Survivors favored LTFU care within the existing consultant/pediatric-led model, followed by nurse-led, distant follow-up and PCP-Led. • Preferences for various models of aftercare were not affected by the level of risk for late effects faced by the patient.
Nathan et al., 2008	Pediatric-Led; Hybrid; PCP	USA and Canada	Risk-Stratified	Cross-sectional survey and analysis of hospital records	8522 Survivors of pediatric cancer	• Despite a significant risk of late effects after cancer therapy, only 31.5% of survivors of CCS received any form of aftercare. • Only 14.6% received care in a cancer centre, with majority of care being received in the community.
Vetsch et al., 2016	Pediatric-Led; PCP	Switzerland	N/A	Questionnaire from Swiss Childhood Cancer Survivor	189 Parents of CCS	• 75% of parents reported that their child attended follow-up. • Of these, 83% reported one or more appointments annually. Majority of the CCS went to pediatric oncologists, while 16% saw their PCP.

Author/Year	**Models of Care Evaluated / Discussed**	**Location**	**Other Features**	**Type of Study**	**Study Participants**	**Conclusions**
						• Most CCS were younger, likely before the transition phase of aftercare. • Researchers recognize that education of CCS and their parents have the opportunity to increase follow-up over the long term.
Nathan et al., 2013	PCP-Led	USA and Canada	Risk-stratified	Cross sectional survey	1124 Family physicians	• 85% preferred working in consultation with a cancer centre. • 48% never or almost never received a treatment summary from the pediatric centre which treated the patient. • Depending on the initial cancer, between 33% and 23 % of PCPs felt very comfortable providing aftercare on their own.
Oeffinger et al., 1998	Survey of Programs Pediatric-Led	USA and Canada	Multidisciplinary	Cross sectional survey	219 Members of Children's Cancer Group / Pediatric Oncology Group	• 53% of responding institutions had a LTFU clinic, 44% followed adult patients. • 93% of clinics were led by a pediatric oncologist. • Barriers identified included: patient uncertainty about the need for follow-up, survivors' unwillingness to attend and locating adult survivors.
Oeffinger et al., 2004	PCP	USA	Hybrid/Primary Care, Risk-stratified	Retrospective cohort study with questionnaire.	9,434 Adult CCS	• PCP provided healthcare for most CCS. • To optimize risk-based care, it is critical that cancer centers and PCP develop methods to communicate effectively.

Author/Year	**Models of Care Evaluated / Discussed**	**Location**	**Other Features**	**Type of Study**	**Study Participants**	**Conclusions**
Parkes et al., 2008	Other Follow-up (Postal follow-up by GP)	UK	N/A	Cross sectional survey	1027 PCP	• The regional cancer program surveyed PCP about 935 survivors who were lost to follow-up. • 88% of PCP responded to the mail survey, providing clinical data around late effects. • Less data was received about socio-economic factors.
Ristovski-Slijepcevic et al, 2009	Survey of Programs; Adult Oncology-Le; Hybrid; PCP-Led; Hybrid	Canada	N/A	Cross sectional survey	17 Pediatric cancer programs	• There is a lot of variation around how aftercare is delivered. • Only 35% programs had access to a follow-up program for survivors after the age of 18. • Other models included: pediatric care, hybrid, and PCP-led.
Sadak et al., 2015	Pediatric-Led	USA	Multidisciplinary	Program description / cross sectional survey	504 Survivors treated at a single centre	• 25% of survivors in the program received their oncology treatment from an outside institution. • With the appropriate healthcare infrastructure, a pediatric cancer survivorship program has the potential to meet the needs of a large survivor population.
Blaaubroek et al., 2007	Hybrid; Pediatric-Led; PCP-led	Netherlands	N/A	Cross sectional survey	358 GP's participated in postgraduate course on late effects in pediatric cancer survivors	• 97% of participants were willing to participate in a hybrid/shared-care model. • 64% felt responsible for caring for CCS. Guidelines and medical history were requested from participants in order to provide appropriate care for CCS.

Author/Year	Models of Care Evaluated / Discussed	Location	Other Features	Type of Study	Study Participants	Conclusions
Casillas et al., 2017	Other Follow-up	USA	N/A	Focus group, key informant interviews, trial of pilot	37 Survivors of childhood cancer survivors	• This text messaging system offered CCS an alternative to regular aftercare. • The system focused on reminders for appointments, late effects, community resources, and messages prompting CCS feedback.
Michel et al., 2017	Survey of physicians providing follow-up care including medical oncologist, pediatricians, GP, pediatric oncologist.	Switzerland	N/A	Survey	183 Physicians. 27 medical oncologists, 122 GP, 13 pediatric oncologist, 21 pediatricians	• Researchers recognized the importance of regular follow-up for CCS despite little insight into physician experiences. • This research was focused on the involvement Swiss physicians, content of their follow-up, problems they faced, and resources needed. • Many physicians, particularly GP's and pediatricians, cared for CCS and suggest the need for standardized care including a standardized MOC.
Lie et al., 2017	Hybrid; PCP-Led; Oncologist-led	Norway	N/A	Focus group interviews	34 CCS of Hodgkin and Non-Hodgkin lymphoma survivors	• The survivors identified their experiences with late effects related care and their preferences for LTFU care. • The shared-care model was suggested to fill the unmet needs of survivors.
Szalda et al., 2016	Adult Oncology-Led; PCP-Led; Hybrid	USA	Hybrid/Shared care	Survey	80 CCS transferred from pediatric survivorship care in	• CCS reported less than adequate engagement with their MOC for survivorship. • Only half of CCS participated in cancer-related follow-up.

Author/Year	Models of Care Evaluated / Discussed	Location	Other Features	Type of Study	Study Participants	Conclusions
						• Of those receiving care, their perception of quality and satisfaction was positive. • The authors suggest that interventions be examined to promote follow-up care.
Signorelli et al., 2017	Pediatric-Led; Nurse-led	Australia and New Zealand	Multidisciplinary	Semi-structured interviews with pediatric medical director and clinical nurse consultant from all LTFU	19 Pediatric medical directors and clinical nurse consultants from LTFU across ANZ	• There is a lack of accepted MOC for CCS across ANZ. • Participants identified limited options for CCS to transition into long-term follow-up. • While they felt that a prescriptive MOC may to too restrictive to best suit CCS, a national MOC with the flexibility to meet individual survivors need may be a useful compromise.

Discussion

We examined the evidence supporting various models of aftercare for CCS. There is some evidence supporting both the pediatric-led and hybrid oncology/PCP models. Phone or mail follow-up was also shown at the very least to maintain contact with some survivors who may otherwise be lost to aftercare. However, much of the research is focused on evaluating individual programs. Few studies examined and evaluated improvements in outcomes associated MOC for CCS. More research is needed, including comparative evaluations of different models, to determine the most effective structure for providing aftercare.

While patients in some studies stated a preference for staying indefinitely with their pediatric care team, many authors recognized that transitioning to an adult provider is more appropriate. It was clear from many of the articles included in this review that transition is a critical component of continued and effective aftercare for CCS (19, 20, 22, 23, 25, 39). We believe that with adequate preparation during the transition phase, a hybrid model/PCP could suit the ongoing needs of many CCS into adulthood. A well-structured transition including effective communication, coordination, health promotion strategies, and engagement of all stakeholders holds the potential to influence the effectiveness of any MOC.

There is a need to further understand aftercare programs that are led by PCPs. It is unclear if aftercare is provided to CCS through family medicine clinics, internists, or

PCPs. It would also be useful to understand if PCP-led models are designed and structured to support CCS or if they are simply default models of care. Nathan et al. identified issues around knowledge and comfort level of PCPs; however, the authors also note that PCP were willing to coordinate and work with other providers to support CCS (28). This opens the opportunity for continuous and coordinated care for CCS. CCS need a certain level of comfort with their providers. Continued care through PCPs while coordinating with pediatric and adult oncologists could allow for both age-appropriate and comfortable aftercare for CCS.

Our review did not examine other features, such as cost, of programs which likely also impacts patient outcomes and satisfaction. Yet, Heirs et al. found that "[i]t was aspects of clinic organisation (e.g., waiting time, length of consultation) rather than the setting or clinic type which seemed to influence patient satisfaction" towards pediatric versus adult care environments (15). Oeffinger found that only around 50% of patients received any type of follow-up care (30). Rebholz et al. reported even lower percentages of CCS receiving care after 10 years in Switzerland (42). Given these findings, ensuring that CCS are receiving appropriate MOC is imperative.

While we found a relatively weak evidence-base for the effectiveness of specific MOC, there is a growing consensus amongst authors and study participants towards a preferred MOC. This model is one in which CCS are risk stratified, with the aftercare of high-risk patients being provided by multidisciplinary LTFU clinics, those at medium to lower risk, being seen by PCP using a shared care model. Survey or phone follow-up may be options for those CCS at the lowest risk. In terms of the shared care component, Singer et al. recommends that a well-organized transition process, provision of a treatment summary and care plan and education with PCPs as components are important elements of a successful shared care model (16). Programs should also ensure access to aftercare for those with limited insurance coverage (43). Hex and Bartlett did not identify any cost-effectiveness studies related to the aftercare for CCS, but they recognize that different models of aftercare have different resource requirements (13).

The main strengths of the review lay in the rigorous methods used to systematically identify the relevant literature, our framework for classifying different MOC and the attention we took in identifying and reviewing articles. Our review had several limitations. Some studies were subject to study design flaws, which could have led to bias in their findings. Potential issues with quality of several included lack of randomization, low sample size, and inherent limitations of observational study designs. Further, our search strategy was designed to be highly sensitive which led to a high number of initial studies. One issue in particular was that there were a number of articles which discussed or offered guidance around models of aftercare, but which did not include new empirical evidence. There is also the risk that individuals included in the study were more invested in the LTFU compared with the average population of CCS. The review was also limited to articles published in English and to full articles indexed in the identified article databases.

Conclusions

With the increase in childhood cancer survival rates and improved knowledge of the late effects of cancer, there is increased demand for appropriate aftercare. In this review, we found evidence, mostly in terms of patient and provider satisfaction, supporting pediatric-led and hybrid oncology/PCP models. However, as found by previous reviews, while there may be an emerging consensus on particular features of a good model of aftercare, there is still a lack of evidence to inform us on how best to structure care (15, 16). Our review illustrates the need to conduct further evaluative research, including evaluations directly comparing different models of aftercare.

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Interventions to Improve the Aftercare of Survivors of Childhood Cancer: A Systematic Review

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Abstract

Purpose: This systematic review summarizes the evidence for the effectiveness of interventions aimed at improving the experiences and outcomes for CCS.

Design: We performed a structured literature search of PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo from 1995 to 2017. Studies were included if they (1) described or evaluated a psychosocial, transition, educational, PA or health behavior modification intervention provided to CCS; (2) presented original empirical research; (3) were published between January 1, 1995 and September 13, 2017; and (4) were full articles, published in English.

Results: Twenty-nine articles met our inclusion criteria. The articles covered five main types of interventions: social skills development, physical activity, workbooks, education, and web-based interventions. Overall, study participants found interventions were useful and showed potential to improve health behaviors for CCS.

Conclusions: Many of the interventions reviewed were helpful to patients and their families; however, most were at a pilot project stage and evidence for their long-term effectiveness was limited across all studies.

Introduction

CCS face significant health risks even after their treatments are completed (1-7). These increased risks result in the need for regular surveillance and many cancer programs have dedicated aftercare programs to provide this care (8). These programs usually focus on both medical issues, e.g., surveillance for recurrence and monitoring of late effects from treatment, and the wider social impacts of being a CCS. Many of these programs have tested interventions to address issues related to health promotion, psychosocial needs, transition, education, PA and other health behavior modification (9, 10). Such interventions can help survivors improve their self-management, self-efficacy and overall QOL. While there are numerous studies examining individual interventions, to our knowledge, no reviews of evidence supporting various interventions have been conducted to date. In this systematic review, we examine the evidence for these interventions to determine which were most promising and could be adopted more widely.

Materials and Methods

Search Strategy and Selection Criteria

We performed a structured literature review of PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo databases. Studies were included if they (1) described or evaluated an intervention provided to a CCS that focused on psychosocial skills, educational attainment, transition from pediatric to adult care, PA or other health behaviors; (2) presented original empirical research; (3) were published between January 1, 1995 and

September 13, 2017; and (4) were full articles, published in English. We excluded evaluations of clinical interventions provided as part of clinical care, evaluations of models / programs of care, accounts of patient need, and diagnostic or assessment tools. The search strategy was reviewed and approved by a university librarian, two experts in conducting systematic reviews, and the study team before being initiated. References for this review were identified through searches of included databases with search terms “pediatric,” “survivor”, “aftercare,” “intervention,” "follow-up," and “cancer.”

Two researchers initially reviewed all titles and removed clearly irrelevant abstracts. Three reviewers then reviewed all remaining abstracts and decisions about exclusion were made by consensus. Final decisions regarding inclusion were made after review of full articles by two researchers. The reference lists of all included articles were also reviewed to identify additional relevant articles. For more detailed information on the search strategy refer to *Appendix A.*

Quality Assessment

Two researchers assessed the methodological quality of all included studies using critical appraisal tools developed by the STROBE statement (11). STROBE offers assessment tools for a variety of study types, which were required because of the variability of study types found in this review. The criteria include questions relating to validity, methodology, analysis and implications of each study.

Data Extraction

The research team developed a standardized data extraction form based on the review objectives. Data fields included a brief description of the study, type of intervention, type of study / evaluation, geographic location where the research was conducted, number of participants, type of participants (i.e., patients, families, or providers), outcomes evaluated and main conclusions. One reviewer extracted study details from all of the articles, with all the completed data abstraction forms being reviewed by another reviewer.

Results

Search Results

Our search identified 10,930 articles. After an initial review of titles, 10,680 articles were excluded because they were either duplicates or they were clearly not relevant to the review. After reviewing the remaining 250 abstracts, a further 120 articles were excluded for not meeting the inclusion criteria. An additional 17 references were then excluded because they were conference abstracts. Of the remaining 102 articles, 74 articles were excluded because they did not meet the inclusion criteria. We included 28 articles from the original search. An additional article was added after review of the references of included articles, resulting in 29 articles included in our review (*Figure 3.2*).

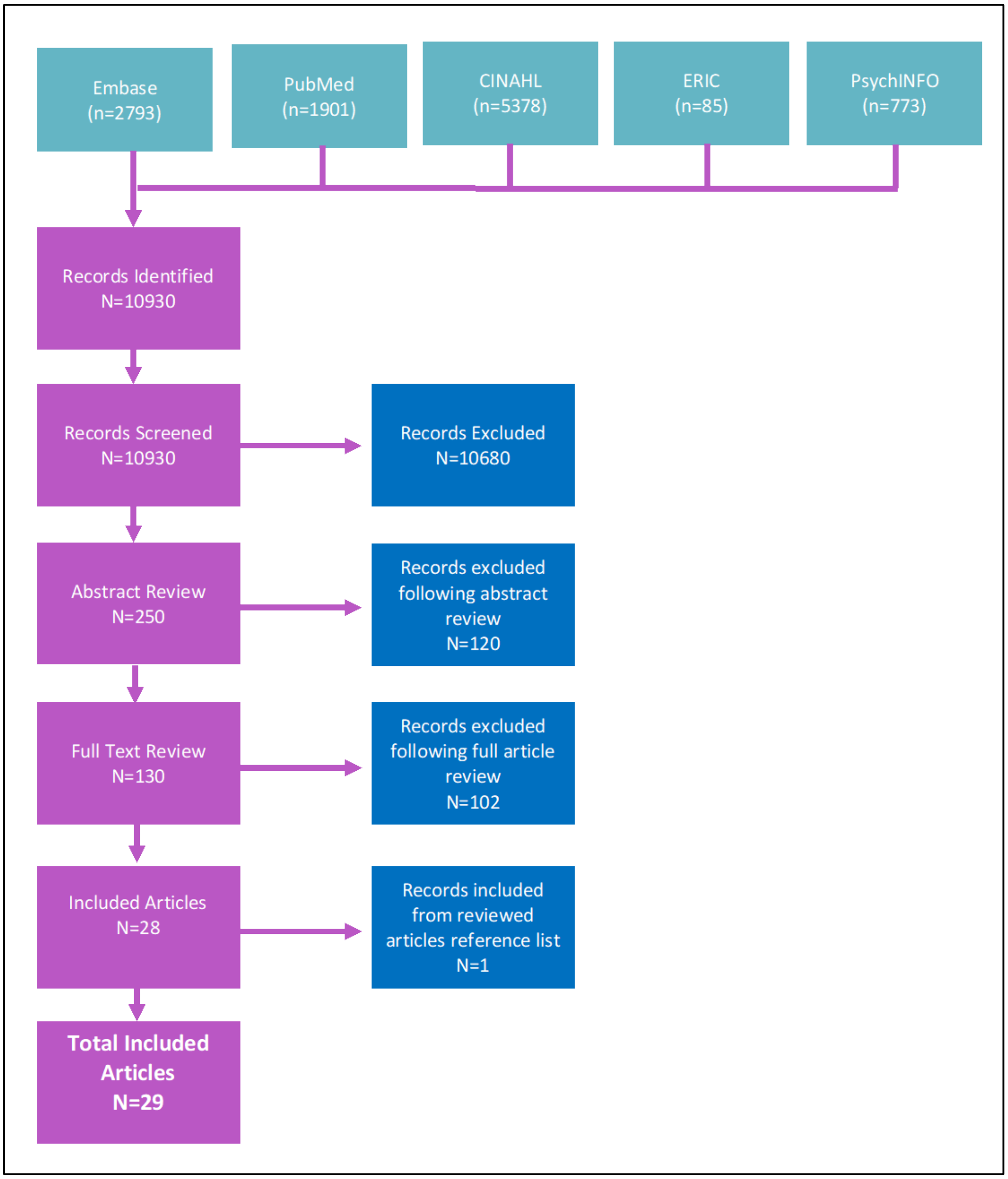

Figure 3.2 *Search Strategy of Interventions for CCS*

Study Characteristics

The 29 included studies are summarized in *Table 3.4*. Most studies (n=21) were published from 2005 onward and were from the USA (n=18). The remainder were from the Netherlands (n=3); UK (n=2); Canada (n=2); Germany (n=2); New Zealand (n=1); and China (n=1). The quality of included studies was generally low and included many observational and descriptive studies. Only seven studies used a randomized design. Furthermore, limited statistical analyses were conducted and many studies were pilot projects focused at a single center.

We present the results in terms of five main types of interventions described in the included articles: social skills development, PA, education, workbooks and web-based. Social skills development is focused specifically on development of social skills such as: cognitive behavioral therapy, self-confidence, social interactions, etc. PA interventions included any fitness-based interventions for CCS. While many of the interventions include aspects of education, this category was focused specifically on counseling on critical aspects of aftercare and not simply providing information. Workbooks included written information provided to CCS related to their aftercare. Finally, web-based interventions included: web sites, mobile applications, and any other electronic sources that were directed towards CCS and their aftercare.

Social Skills Development Interventions

We found seven studies that examined interventions that supported social skills of survivors. *Surviving Cancer Competently Intervention Program (SCCIP*) was a 1-day intervention developed at the *Children's Hospital of Philadelphia* (12). The intervention targeted anxiety, beliefs about cancer and its treatment, social support and family communication with the aim of reducing symptoms of PTSD in CCS and their families. SCCIP was comprised of four family and larger group sessions, and utilized a cognitive-behavioral and family-therapy approach. This intervention targeted CCS, their parents and siblings. Participants were positive about the program; however, Kazak et al. found no statistically significant differences between CCS participating in the intervention compared to controls in terms of anxiety, PTSD, and event impact scales (12). Kazak et al. later completed a randomized clinical wait-list trial for the SCCIP program and found that brief interventions reduced PTSS in this population (13). Similar to the pilot, results suggested that interventions to address PTSD were important for CCS and their families.

Santacroce et al. evaluated a social development intervention that focused on a telephone directed coping skills training (14). They found in-person, LTFU supported by telephone-delivered psychosocial care was a useful way to provide care to CCS and their parents (14).

Schwartz et al. evaluated a three-day intervention called "Moving On," focused on psychosocial improvements for CCS (15). The intervention included discussions, presentations, and group activities. Researchers employed a pretest-posttest design to examine response shift due to treatment evaluation and a case-control design to examine differences between survivors and non-survivors in reported QOL. While the intervention yielded positive short-term results, these improvements reverted closer to those of an age-matched group of CCS controls after 3 months (15).

Barrera et al. studied a group intervention for brain tumor survivors who previously reported social concerns. The intervention aimed to improve key social skills (e.g., social initiation, cooperation, managing bullying, conflict resolution, building empathy and self-confidence) (16). Each session focused on a single social skill and included group discussions supervised by a psychologist, structured social interactions, breaks and homework. The analysis showed good recruitment and retention throughout the program and significant improvement after the intervention from parent reports on self-control, social skills and QOL (16).

Schulte et al. piloted a very similar intervention for CCS of central nervous system cancer in Alberta, Canada (17). The intervention focused on six skills (friendship making, cooperation, managing bullying, conflict resolution, empathy and assertion) that were based on previous intervention work. Researchers found some improvements in parent-reported social skills (17).

Boston Children's hospital introduced *Success Through Education, Psychosocial support, and Socialization Program (STEPS)* (18). This program provided survivors of pediatric brain tumors a structured intervention for socialization and psychosocial support. The researchers found that parents of individuals who received the intervention reported increased confidence in their child, despite no increased social interaction beyond that with others CCS who participated in the STEPS group (18).

PA Interventions

Six articles studied interventions focused on increasing PA. Several studies employed PA tracking to enhance physical activity. Mendoza et al. evaluated a 10-week intervention using a FitBit to track PA and a peer-based Facebook support group (19). Qualitative results indicate that this invention was acceptable for CCS; however, the quantitative results indicated no significant changes in moderate to vigorous PA. There were also slight differences in improved QOL and motivation for PA reported by the study participants (19).

Le et al. evaluated a home-based exercise intervention using *Fitbit One* exercise trackers (20). The intervention consisted of wearing a "Fitbit One" device for 6 months. The researchers completed testing at baseline and follow-up examined self-reported surveys, an accelerometer for 7 days and VO2 maximum test through cardiac stress test. This intervention assessed feasibility, impact on activity levels and physical fitness, barriers, preferences, and beliefs regarding PA of CCS. Despite being a pilot study, this research showed high participant retention (79%), receptivity and belief of utility (20). Results illustrated an increase in total weekly moderate to vigorous activity and an increase in VO2 max was also observed. Despite no statistically significant results, this intervention showed to be a motivational tool to modestly improve physical activity.

Li et al. studied an adventure and education based experience for CCS who were at least 6 months off treatment, between ages 9-16 years, and were not involved in regular PA (21). Researchers used a single-blind randomized control trial (RCT) with a two-group pretest and repeated post-test (21). They found CCS who participated in the integrated program reported significantly higher levels of PA and self-efficacy when compared to the control group (21). The authors suggested that education alone was insufficient to change behavior, but integrated health education and PA training may have the potential to change CCS behavior.

Ruble et al. studied a 5-day camp followed by 5 monthly follow-up sessions which took place over a 6 month period (22). The outcome measure of the study included feasibility, change in amount of moderate to vigorous activity, and self-efficacy. Participants who were randomized into the intervention group completed a 5-day PA day camp. As expected, individuals who participated in the intervention had higher rates of moderate to vigorous activity(22).

Takken et al. developed “FITstrong,” a 12 week training program focused on aerobic and strength exercises (23). The goals of the intervention were to develop a home-based PA program for CCS and examine its efficacy and feasibility (23). Researchers found a number of challenges with their PA program, including issues with motivation, sustainability, and education of the importance of PA and participation (23). Additionally, only participants who completed the exercise program completed the evaluative questionnaire (23).

Wynn et al. created an outdoor wilderness adventure therapy intervention to address late effects and enhance self-esteem, self-discovery and life skills in adolescents (24). Participants completed a 14-item resilience questionnaire and a group cohesion measure but results were limited by low number of participants (24). Overall, improvements in resilience were noticed in CCS and the authors concluded that the intervention is worth exploring further.

Educational Interventions

Eight of the articles evaluated educational interventions. Park et al. developed and evaluated a national (USA) peer-delivered telephone smoking cessation intervention for adult CCS who smoke (25). The intervention included 6 counseling calls tailored to an individual's circumstances and provided supportive materials (e.g., the provision of nicotine replacement therapy). Retention was an issue as only 41.5% of study participants attended at least 5 calls. 20% of patients who completed the intervention reported having stopped smoking after 12 months, compared to only 3% of patients who did not participate in any calls (25).

Hudson et al. evaluated an intervention designed to improve health related QOL (26). The researchers randomized patients into two groups. One group received the intervention, which was comprised of late-effects risk counseling, provision of a clinical summary, health goal commitment to practice health related behaviors, health behavior training and included a telephone follow up 3 and 6 months after the intervention. Preliminary results of this pilot intervention showed that educational interventions in a specialty clinic for CCS can provide health-related quality support to participants (26). In 2002, Hudson et al. evaluated an intervention designed to improve knowledge of late effects and treatment-related health risk using a RCT (27). Health behaviors were measured using a 38-item Health Protective Behavior Questionnaire (27). Based on self-reported health practices, this intervention improved QOL for survivors in a LTFU clinic when compared to their standard care (minimal duty of care required for patients).

Patel et al. piloted an intervention directed towards improving parenting skills (i.e., parent education, efficacy, behaviors and child academic scores) and CCS education functioning (28). Outcome measures were evaluated using pre- and post-measures from the Wechsler Individual Achievement Test-II and the Scholl Motivation and Learning Strategies Inventory (28). Only modest effects on outcomes including level of knowledge, self-efficacy and educational success where identified (28).

Bava et al. completed a parent-directed pilot intervention to improve academic support for school-aged survivors of childhood acute lymphoblastic leukemia (ALL) (29). Children-parent dyads participated in the intervention. The intervention first assessed cognitive, academic and socio-emotional functioning by standardized measures. The dyads also attended a feedback session that included the standard intervention to receive psycho-educational assessment results and general recommendations for academic support. The outcome measures looked at assessing the feasibility and determine predictors of participation in an academic intervention for this population. The authors conclude that this intervention provides competent assessment and academic support for CCS (29).

Maurice-Stam et al. evaluated *OK Onco*, a psycho-educational group intervention in the Netherlands(30). The intervention is focused on emotional support improving adaptation, and specific skills and reduction of physical symptoms by behavior change. Eleven participating CCS and their parents completed questionnaires about the disease-related skills that were taught in the intervention, before the start of the intervention and 0-4 weeks after the intervention. The analysis from two disease-related skills questionnaires showed positive outcomes for most disease related skills, such as relaxation, social competence and positive thinking (30).

Camp *Mak-a-Dream* was a 4-day camp with CCS to enhance their advocacy skills. Upon completion of camp, the researchers had the participants complete a brief questionnaire. The researchers found that participants appreciated the opportunity to share experience and provide peer support and described the intervention to be comforting and healing (31).

Rothbart-Mayer et al. evaluated *Reunion Day,* which was created to provide a supportive forum, examine QOL, and assess patient needs for CCS and their families (32). CCS offered recommendations to enhance their healthcare, including the need for transitional support and enhanced education of long-term needs for CCS (32). CCS reported that these educational interventions were able to help them build relationships and share their stories while modestly improving their ability to acquire information about their cancer.

Workbooks

Three articles evaluated the use of information booklets for patients as a means of better informing and motivating CCS. Blacklay et al. evaluated the effectiveness of an information booklet developed by the Children's Cancer Study Group's Late Effects Group in the United Kingdom for young adult CCS to increase knowledge and influence health-related behaviour (33). This booklet included information on health issues related to being a CCS, the type of treatments received, reasons for continued follow-up, the impact of CCS on various life issues and emotional issues related to being a CCS. Future versions of the booklet plan to include additional information focused on a patient's specific cancer and treatment. Blacklay et al. found that there was a high level of patient satisfaction with the booklet, and greater appreciation of certain health risks and for the need for follow-up reported by patients, as determined by a telephone interview one week after receiving the booklet. Absolom et al. examined the use of a similar booklet in South Yorkshire (UK) aimed at adolescent

CCS (34). Absolom et al. found that CCS who read their booklet had “a more positive attitude to [follow-up] clinic.”

Bashore et al. evaluated an interactive transition workbook (35). This workbook included health information, including type of cancer, treatments, insurance information and information about HCP. Patients completed this workbook over a six-month period with staff support during their period of transition out of adult care. Bashore et al. found that a third of patients did not complete the work plan or were lost to study follow-up, but that there was a high level of patient satisfaction for those who did complete the workbook (35).

Web-based Interventions

Five web-based interventions were identified. Cancer SurvivorLink is a web-based data-sharing application developed in Georgia (U.S.), which allows patients to share documents and information with their provider (36). It serves as an electronic health record, which is created and managed by the CCS or their relative. The evaluation mostly focused on the recruitment for Cancer SurvivorLink. The application initially showed a high level of uptake after employing a variety of recruitment strategies. However, as commitments on patients increased (e.g., creating a personal record, uploading documents and sharing documents), participation declined significantly, with only 21% of participants ultimately sharing their information with a HCP. Attendance at a survivor clinic was the greatest predictor associated with creating a

SurvivorLink account. Researchers suggest more education around the need for follow-up is required to increase the uptake of this type of shared clinical care intervention (36).

In 2012, Knijnenburg et al. examined a Dutch website that provides information on late effects and other medical issues to CCS and their families (37). The website is designed to allow the user to search through types of cancer and treatment to identify information about the specific late effects that a CCS may face. The evaluation focused on CCS information needs and examined the usability, layout and satisfaction with the website. Participants rated the usability of the site as 72.5 out of 100, and the content 3.7 out of five. Respondents indicated that they would have preferred even more detailed and scientific information about late effects to be provided, for the search tool to allow for even more focused late effects information for specific patients and for the site to be regularly updated to encourage people to return to it (37).

Kunin-Batson et al. piloted a similar website, which aims to provide tailored health and follow-up information to CCS (38). They evaluated the impact access to the website would have versus standard physician counseling. However, only 46% of the CCS randomized to the website arm of the trial actually visited the website. Of those that did visit the site, only 33% visited the site more than once, with the average time on the site being 13 minutes. Of those who visited the website, 71% reported being "satisfied" or "very satisfied" with it.

Onco-STEP is an internet-based psychotherapeutic intervention developed in Germany to help reduce posttraumatic stress symptoms and help patients cope with cancer related-fears (39). Onco-STEP used 10 writing assignments completed by CCS and then reviewed with feedback provided by a therapist in reference to a structure treatment manual developed for the intervention. The evaluation showed that 80% of the 20 participants were satisfied with the models and the feasibility of providing internet-based psychotherapeutic to CCS, however, researchers acknowledge the need for further evaluation.

Kock et al. developed a mobile application, CancerLateFX, to manage and reduce the risk of late effects and remind German CCS of follow-up appointments (40). CCS and their relatives completed questionnaires which evaluated the application in seven areas: suitability, descriptiveness, controllability, conformity, error robustness, customizability and suitability for learning. CCS and their relatives felt positively about the mobile application and its ability to hope manage aftercare visits, but few other details about the evaluation were provided in the article.

Table 3.4 *Characteristics of Included Studies*

Author, Year, Location	**Type of Intervention**	**Intervention Summary**	**Study Type**	**Participants**	**Outcome Measures**	**Conclusions from articles**
Barrera et al., 2009 Canada	Social Skills	Social skills group intervention, which includes eight two-hour sessions	Pre & post intervention assessment	32 CCS	Social skills, quality of life, self-efficacy, and depression	The intervention is feasible and provides preliminary support for the efficacy of the program.
Kazak et al., 1999 USA	Social Skills	Cognitive-behavioural and family therapy intervention	Pre & post intervention assessment	19 CCS and their families	Distress, family functioning and development, PTSS, and anxiety	The data was supportive of SCCIP's effectiveness and feasibility.
Kazak et al., 2004 USA	Social Skills	Four-session, one-day intervention, integrating cognitive-behavioural and family therapy. Aimed to reduce PTSD	Randomized control trial with wait-list control	150 CCS and their families	PTSS outcomes and anxiety	Brief interventions can reduce PTSS in this population. Family support was found to be important to success.
Santacroce et al., 2010 USA	Social Skills	The HEROS PLUS CST is a 7-session telephone intervention directed towards coping skills training and psychosocial care for CCS and parents	Randomized clinical trail	15 CCS and 16 parents of CCS	Uncertainty, anxiety, port-trauma, benefit finding, and health promotion behaviour	HEROS PLUS CST has clinical relevance through delivering telephone psychosocial care coupled with in-person long-term care as a means to provide integrated support for CCS.
Liptak et al., 2016 USA	Social Skills	The STEPS was developed to provide adolescent and young adult survivors of pediatric brain tumors structured opportunities for socialization and psychosocial support.	Mixed methods using phenomenological approach	19 survivors (14 females, 5 males) and 18 care-givers	Report of their beliefs and actions	Significant social isolation that was compounded by medical late effects. Survivors perceived social support and acceptance from interactions with peers who have similar medical backgrounds as a key aspect of the group experience. Parents reported increased social confidence among survivors, although they did not report that social gains generalized beyond the group setting. Interventions to promote the transfer of specific social skills are needed.
Schulte et al., 2014 Canada	Social Skills	Social skills intervention program	Pre & post intervention	15 CCS of CNS tumours in intervention	Social skills, social problems, and social functioning	The intervention showed improvements in parent, teacher, and survivor social

Author, Year, Location	**Type of Intervention**	**Intervention Summary**	**Study Type**	**Participants**	**Outcome Measures**	**Conclusions from articles**
			assessment with control group	group, 12 individuals in control group		adjustment scores when compared to control group.
Schwartz et al., 1999 USA	Social Skills	Psychosocial intervention focused on QOL for CCS	Pre-post evaluation & case-control design	22 CCS and 54 healthy controls	Health related quality of life, psychological well-being, and response shift	The intervention regulated survivor's conceptualization of QOL so it was similar to age-matched control. Future research should consider response shift in randomized treatment evaluation.
Knijnenburg et al., 2012 Netherlands	Web-based	Website for CCS	Questionnaire	55 CCS, 43 parents of CCS	Medical decision style, usability and content of the website	Respondents were satisfied with the usability and the contents of a website. The effects on survivorship care were not evaluated.
Kock et al., 2015 Germany	Web-based	Mobile application to manage and minimize the risk of late effects	Questionnaire	13 CCS, 9 relatives	Components of mobile application	No evidence of the compliance is provided. The app needs to be subjected to a long-term evaluation.
Seitz et al., 2014 Germany	Web-based	Web-based psychotherapeutic intervention intended to reduce PTSD and provide support and coping mechanisms	Questionnaire	20 CCS	Socio-economic background, cancer history, psychosocial services, suicidality, and PTSS	CCS accepted this web-based psychotherapeutic intervention.
Williamson et al., 2014 USA	Web-based	Patient-controlled communication tool where survivors can electronically store and share documents with healthcare providers	Evaluation of intervention	275 CCS and parents	Registration, creation of personal health record, and uploading documents	Attendance at a survivor clinic was the biggest predictor of registering and using SurvivorLink. Survivors advocate for their aftercare and this intervention provides support for their decision-making.
Kunin-Batson et al., 2016 USA	Web-based	Researchers evaluated the impact to a CCS website versus standard physician counselling	RCT	52 CCS	Surveys measuring cancer knowledge, health locus of control and psychosocial well-being	Utilization of the website was low; therefore, it was difficult to draw conclusions about efficacy. Providing personalized information through the web was not more effective than standard of care at improving cancer knowledge.
Li et al., 2013 China	PA	Four-day integrated adventure training and health education program	RCT	71 CCS	PA levels, self-efficacy, and quality of life	The intervention was effective in promoting regular PA among CCS.

Author, Year, Location	Type of Intervention	Intervention Summary	Study Type	Participants	Outcome Measures	Conclusions from articles
		to promote regular PA in CCS.				
Le et al., 2016 USA	PA	Novel home-based exercise intervention with Fitbit One exercise trackers	Pre & post intervention assessment	19 CCS	Feasibility, impact on activity levels and physical fitness, and barriers, preferences, and beliefs regarding PA	This pilot study had high participant retention, receptivity, and belief of utility. Further studies should use a large sample and evaluate sustainability of this intervention.
Mendoza et al., 2017 USA	PA	10-week intervention using PA tracking (Fitbit Flex) and a peer-based virtual support group using Facebook.	Two arm, unblended, randomized control trial	59 CCS	PA levels, psychological needs satisfaction, motivation, PedsQL generic core, PedsQL cancer module	This intervention was feasible for CCS and some modest differences were found in QOL and motivation for PA.
Ruble et al., 2016 USA	PA	5-day camp, followed by 5 monthly camp reunions.	Randomized, control group design.	9 CCS and 10 controls	Measures of feasibility, change in percentage of awake time spent in moderate to vigorous activity, self-efficacy scores, and correlations in moderate to vigorous activity and self-efficacy	Increases in moderate to vigorous activity are seen in CCS who participate in a group intervention and this includes support of self-efficacy.
Takken et al., 2009 Netherlands	PA	Twelve-week exercise program for CCS	Evaluation of intervention	9 CCS	Efficacy, feasibility, muscle strength, exercise capacity, functional mobility, and fatigue	Only 4 participants were able to complete the program. A balance between age disease stage; variety of exercises; location; motivation, should be considered in order to lead to better adherence.
Wynn et al., 2012 New Zealand	PA	Eight-day wilderness adventure therapy intervention to enhance self-esteem, self-discovery and life-skills	Pre & post intervention assessment	5 CCS and 5 support staff	Resilience, cohesion, autonomy, and social support	Despite limited statistical evidence, participants felt the intervention was a growth-enhancing experience.
Bava et al., 2016 USA	Educational	A parent-directed intervention to increase academic support for school-aged survivors of childhood ALL	Non-randomized, prospective cohort pilot study	49 participants	Assess the feasibility and preliminary efficacy of a clinical service routinely provided and determine predictors of participation	The feasibility needs to be assessed; however, this approach enables pediatric cancer treatment centers to provide culturally competent assessment and academic support.

Author, Year, Location	**Type of Intervention**	**Intervention Summary**	**Study Type**	**Participants**	**Outcome Measures**	**Conclusions from articles**
Hudson et al., 1999 USA	Educational	Health promotion intervention to improve QOL for survivors in LTFU clinic	Prospective, randomized controlled feasibility study	266 families	Baseline knowledge, burden of performing the intervention, quality of life, and health goal commitments	This study supports the feasibility of an educational intervention in a specialty clinic dedicated to monitoring long-term CCS.
Hudson et al., 2002 USA	Educational	An intervention designed to improve knowledge of late effects and treatment-related health risk	RCT	251 CCS	Health behaviors were measured using a 38-item Health Protective Behavior Questionnaire	Based on self-reported measures, this intervention improved QOL for survivors in LTFU clinic when compared to their standard care.
Maurice-Stam et al., 2009 Netherlands	Educational	Psycho-educational group intervention aimed to empower CCS	Questionnaire and focus group	11 CCS and their parents	Survivorship related skills and effect of intervention	Researchers found positive outcomes with respect to disease-related skills, social competence, and positive thinking.
Park et al., 2006 USA	Educational	Telephone-based, per-delivered smoking cessation intervention for CCS	Randomized trial	398 CCS (current or former smokers)	Demographics, cancer history, smoking outcomes, and psychosocial factors	Peer-delivered, telephone counselling is an effective way to reduce adult survivors that smoke.
Patel et al., 2014 USA	Educational	Intervention to improve parenting skills and child's educational functioning for CCS with neurobehavioral late effects	Randomized pre & post assessment	44 CCS and their parents	Parent knowledge, efficacy, learning behaviours, and child academic scores	This intervention is feasible and effective for improving targeted parenting outcomes and child academic outcomes.
Rothbart-Mayer et al., 2000 USA	Educational	Workshop for CCS and family	Descriptive	40 CCS and 44 family members	Health issues, psychosocial issues, and post-treatment needs	The workshop provided a preliminary assessment of survivors' needs (QOL, education, and post-treatment needs).
Zebrack et al., 2006 USA	Educational	Four-day retreat that provides education and support	Pre & post intervention assessment	35 CCS	Issues of survivorship care, cancer education, tools for self-advocacy, and support for survivors	No formal analysis was conducted, but participants were offered experiences that may promote successful achievement of age-appropriate developmental tasks.
Bashore et al., 2016 USA	Workbook	Transition workbook	Mixed methods design	30 CCS	Determine the most helpful sections of the workbook, the time needed to complete the work-book, staff time assisting	This study identified the need for collaborative transition programs using methods of addressing transition readiness and evaluation.

Author, Year, Location	Type of Intervention	Intervention Summary	Study Type	Participants	Outcome Measures	Conclusions from articles
					participants in completing the workbook, and the impact of completing the workbook on measures of worry and transition readiness.	
Absolom et al. et al., 2004 United Kingdom	Workbook	Workbook to increase knowledge surrounding health promotion for CCS who still attend follow-up	Workbook	48 CCS	Increased knowledge and awareness of cancer and its treatment, education around health risks with respect to cancer treatments and promotion of healthy lifestyles.	CCS reported a more positive attitude towards aftercare clinics after completing workbook. They also reported being ready for health promotion behaviors.
Blacklay et al., 1998 United Kingdom	Workbook	Workbook for information about healthy lifestyle, need for long-term follow-up, employment, and insurance for CCS	Workbook and interview	50 CCS	Outcome measures include understanding of illness, treatment, impact of diagnosis, and preferred information	CCS were interested in receiving more written information. They also learned about the importance of health practices including the risks of sun bathing and the need for long-term follow-up. Written information should be provided to CCS during aftercare.

Discussion

This review identified 29 studies that used patient and family-focused interventions to enhance health behaviors of CCS. These interventions aimed to augment existing aftercare programs and offered survivors the opportunity to address a wide range of potential physical, educational and psychosocial issues. Given the breadth of this review, the relatively limited number of studies identified indicates that this important area of aftercare remains understudied. Many of the articles were limited to pilot projects or were focused on single-center evaluations with small study populations. Future research should aim to be conducted across multiple centers. Such research is essential to better evaluate these types of interventions and identify those which warrant more wide scale adoption.

Methodological issues were common, including small sample size, minimal follow-up time, observational study design, lack of validated outcome measures and pilot designs. Despite these limitations, some of the interventions identified are promising for improving survivor care. Interventions focused on social skill development, like SCCIP (13), the Moving-On program (15), or the program developed by Barrera et al. (16), while not demonstrating clear results, did have high levels of patient and family satisfaction. Similarly, while there was limited evidence of long-term effectiveness, participants indicated that the physical exercise interventions reviewed were important developmental experiences. Studies also demonstrated that CCS who participated in PA interventions had higher levels of self-efficacy, which may help CCS commit to a healthier lifestyle. Workbooks also show the potential of increasing patients' knowledge, improving attitudes towards healthy behaviours and reducing some of the anxiety around the transition into adult care.

We identified five studies that focused on web-based interventions. The literature suggests that web-based interventions show improvement in knowledge and/or behavior change (41) and have the potential to improve health outcomes of CCS (42). Given that accessibility is one reason patients avoid aftercare, these types of interventions may help CCS to remain engaged in aftercare (42, 43). Web-based interventions have the potential to be more effective when augmented with additional methods of communicating with participants, i.e., SMS or text messages (43). Further, the use of cellular devices and mobile applications offer ease and functionality to their users (41, 42, 44) and mobile applications have been shown to modify human behaviors(43). Interventions of this type may help with awareness of survivorship information, follow-up practices and survivorship autonomy. As with the workbooks, it is key to ensure the level of uptake of these interventions warrants the amount of effort to develop them. The approach taken by Knijnenburg et al. and Liptak et al. to focus interventions at a national level could be used to help address this issue (18, 37).

Several of the included articles focused on both survivors and their immediate family (12, 16, 28-30, 36, 40). The literature suggests that social support and family cohesion are important components of successful aftercare (45). Overall, survivors and family members reported being satisfied with most interventions. Social adjustments were made in areas such as friendship, cooperation, QOL and social support through group skills programs, a 1-day intervention session, a psychosocial intervention and group psych-educational intervention (12, 15-17, 30, 37, 46). No study included in our review addressed the needs of CCS who do not have strong family support structures, indicating an opportunity to design and study interventions targeted at this critical group.

There were other gaps in the literature identified in our review. Transitioning effectively from pediatric to adult care is imperative to monitoring and managing potential late effects throughout a CCS life, and successful transition practices are associated with improved health outcomes (47). Despite this, our review identified only one intervention that directly supported transition (35). While the interventions we examined have shown modest improvement in cancer aftercare behaviors, it remains to be seen if they are economically feasible or cost-effective on a larger scale or the long term, as no studies examined this aspect of the interventions. Finally, many of the interventions targeted CCS who already regularly attend follow-up clinics. An important group of CCS, who are likely at higher risk for adverse health outcomes, are those lost to follow-up.

Our review had a number of limitations. We restricted our review to peer-reviewed published studies, and we did not search for grey literature in this field. It is possible that other settings have completed evaluations of interventions for CCS but that the results have not been published in the peer reviewed literature, or that there are interventions occurring that have not been formally evaluated. Our search strategy was designed to be very inclusive, but this meant that many articles in our search were ultimately not relevant. Additionally, our broad search strategy led to a mixed group of studies with different study designs, populations and outcome measures. The review was also limited to articles published in English and to full articles indexed in the identified article databases. These search restrictions meant that some research may not have been identified through our review.

Conclusions

The interventions reviewed were shown to be supportive for patients and their families, however, the evidence supporting their role in long-term behavioral change or improved health outcomes is limited. Evaluating the quality of interventions was challenging due to the heterogeneity of study designs, lack of validated outcome measures and other factors. Research focusing on interventions to improve the transition of care for CCS from pediatric to adult care is essential, as is research that examines interventions designed to improve the self-efficacy of CCS. The accessibility, ease of use and low cost of web-based interventions that use mobile technology makes them a promising venture for future research and care provision. Overall, there is insufficient evidence to recommend any particular intervention. However, given the possible impacts interventions can have on CCS, it is important that cancer programs continue to develop innovative ways to better support CCS.

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Chapter 4

Transition and Aftercare for Survivors of Pediatric Cancer in Newfoundland and Labrador

Introduction and Overview

The research from Chapter 3 identified that transitions from pediatric to adult care were largely under-represented in the referenced studies. As such, we used a qualitative research design, specifically, a Case Study design to explore this area further. This study describes how transition from pediatric to adult care occurs for CCS in Newfoundland, Canada. Moreover, this chapter highlights the barriers that CCS face in their transition. Additional information on the methods of this study can be found in *Appendix A*.

This qualitative research aligns with element two of the WHO framework. This case study is the second part of the situational assessment. This analysis is the foundation for establishing how current processes of transition are occurring and planning for new interventions. The WHO framework suggests a comprehensive understanding of the health system. This includes current health care structures, obstacles, opportunity for improvement, health goals, and priorities. The aim of this research is to gain a comprehensive understanding of current transition and aftercare processes and provide insight into future directions for interventions.

Co-authorship Statement

Transition and Aftercare for Survivors of Pediatric Cancer in a Canadian Province: A Qualitative Study

- Devonne Ryan developed the research design, supervised by Dr. Roger Chafe and Dr. Paul Moorehead.
- Devonne Ryan recruited participants assisted and supervised by Dr. Paul Moorehead.
- Devonne Ryan drafted the manuscript.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead edited and critically appraised the manuscript.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead read and approved the final manuscript.

Transition and Aftercare for Survivors of Pediatric Cancer in Newfoundland and Labrador: A Qualitative Study

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Abstract

Background: Childhood cancer survivors (CCS) face increased risks during the period when they leave pediatric centres and transition into adult-focused aftercare. We examine CCS's experiences entering adult-focused aftercare across Newfoundland and Labrador (NL) to better understand current transition practices, barriers to transition, and identify opportunities for improving care.

Methods: We conducted both in-person and telephone semi-structured interviews. CCS who recently transitioned out of pediatric care and healthcare providers (HCP) who provide care for CCS in NL were identified using purposive sampling. Participants were interviewed between July 2017 and March 2019. Data was analyzed using both qualitative description and thematic analysis.

Results: Five CCS, five adult or pediatric oncologists, three allied health professionals, and one family physician were interviewed. No structured transition process for CCS was identified in the province. All CCS reported receiving aftercare through a children's oncology program, while only two reported receiving any form of aftercare in an adult setting. The barriers to improved transition included: added challenges for survivors in rural areas, changes in the services available in adult-focused aftercare, challenges navigating the adult system and lack of education on transition and aftercare.

Interpretation: We found that there was little preparation and disruptions in aftercare related to CCS's transition into adult care. Programs serving CCS should aim to make these transitions more standardized and better supported, e.g., through the development of context appropriate educational resources.

Keywords: Cancer survivorship; childhood cancer; aftercare; transitions

Introduction

Advances in the treatment of childhood cancer have led to dramatic increases in the number of patients who survive their cancer and live well into adulthood (1-5). Yet approximately two thirds of these childhood cancer survivors (CCS) will be negatively impacted by their past cancer or from late effects from its treatment (6, 7). Because of these elevated health risks, it is recommended that CCS be followed throughout their lives by appropriately trained Health Care Providers (HCP) (6, 8-10). This period of surveillance care is called follow-up care or aftercare. In *Figure 4.1*, we present a model we developed based on our previous work that illustrates the typical disease-treatment progression for a CCS to highlight the place for transition and aftercare(5, 11). After the onset of cancer and its symptoms, the cancer is diagnosed and treated. After treatment ends and the patient has no active cancer, there remains a life-long risk of cancer recurrence and negative impacts from receiving cancer treatment. Aftercare, which is the follow-up care received after active disease treatment ends, is dedicated to monitoring and managing these possible late effects. After treatment and aftercare in pediatric care settings, many CCS will transition and continue their aftercare in an adult-focused health care setting (12). A key point for ensuring the continuance of aftercare is when CCS become adults and they usually leave pediatric programs and transition into the adult-focused health care system.

Despite clear health benefits, including continued care to manage late effects there are many challenges to maintaining optimal patient care during this transition into adult care (13-15). While it is recognized that the transition into adult care is a critical period for CCS, we found few rigorous, detailed accounts of how this transition occurs and barriers to transition during a recent systematic review of aftercare programs (5, 16). Similarly, there were a limited number of interventions evaluated that were focused on supporting the transition of CCS, especially for interventions not limited to a single aftercare program (13, 17, 18). We explored current transition practices in the Canadian province of Newfoundland and Labrador (NL). We focused on NL for several reasons. It has relatively small population (521,542) that covers a large geographic area (405,720 km^2) with many rural and remote communities(19). NL only has one pediatric cancer program, located in its capital city, which treats patients from across the province. As like the rest of Canada, NL has public health insurance which provides universal coverage for pediatric and adult follow-up cancer care. The pediatric oncology program at the Janeway Children's Health and Rehabilitation Centre (often referred to as the Janeway) manages between 12 to 18 new pediatric cancer cases per year. Given the small size of the program, its HCP often develop lasting relationships with their patients and the program has high rates of retention within its pediatric aftercare program. Still it is unclear how and to whom survivors' transition once they entered the adult-focused health care system across the province. We aim to use this increased understanding of current transition processes to identify contextually appropriate interventions focused on improving transition and aftercare for these survivors. Our objective is to conduct an in-depth examination of current transition practices across NL to better understand current transition practices, to identify barriers to transition, and find opportunities for improving survivorship care.

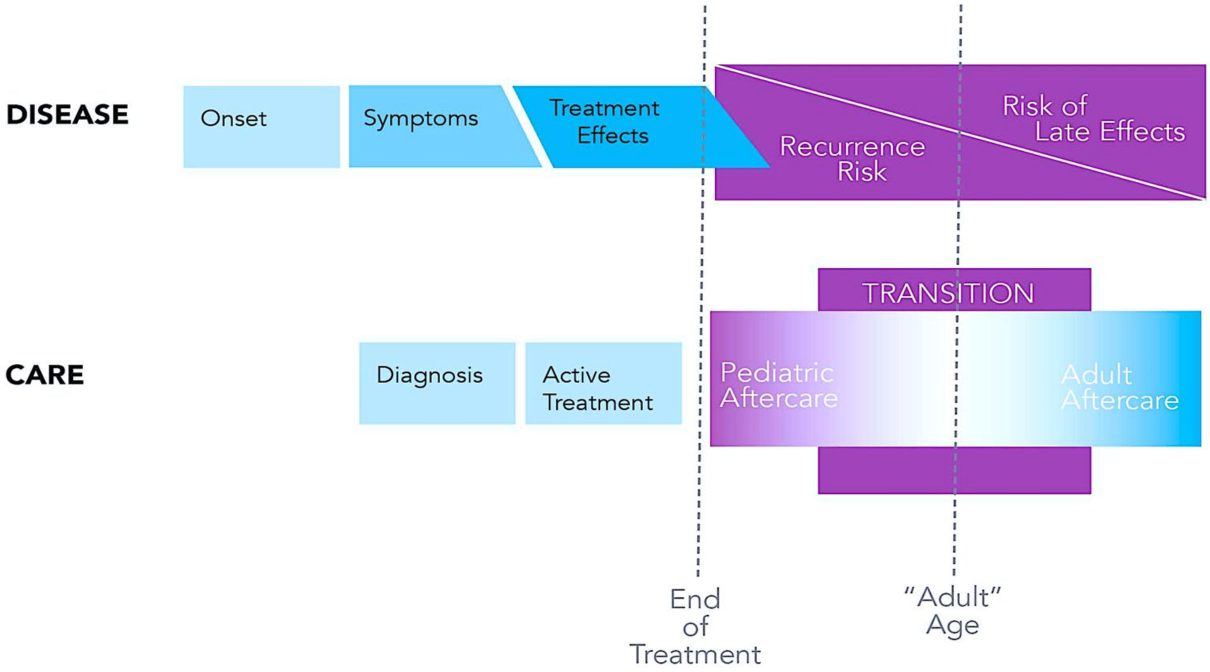

Figure 4.1 *Disease - Treatment Progression*

Methods

Study Design

We used a qualitative research design based on key informant interviews, following the *Consolidated criteria for reporting qualitative research* (COREQ) (20).

Study Setting

This study was completed in NL between July 2017 and March 2019. CCS who participated completed their care at the Janeway Children's Health and Rehabilitation Centre located in St. John’s, NL. Pediatric HCP interviewed all worked at the Janeway, which provides pediatric oncology services for the entire province. The adult-focused HCP interviewed all work at the Dr. H. Bliss Murphy Cancer Centre or the Health Science Centre, both located in St. John’s, NL.

Study Participants

Study participants included pediatric-focused HCP, adult-focused HCP and CCS who completed their care in the Janeway pediatric oncology program. HCP and CCS were identified and recruited using purposive sampling (21, 22). All HCP who were recruited were directly involved in the care of CCS. The Janeway Pediatric oncology program consists of three full-time pediatric oncologists, who were all invited to participant. Other HCPs who support CCS, including a physiotherapist, a psychologist, an oncology nurse, a dietitian and social workers, were also invited. These HCP were identified by the Janeway pediatric oncology program and were recruited by e-mail. In total, invitations were sent to 12 HCP. 20 CCS who were 1) diagnosed with cancer before 18 years of age; 2) treated at the Janeway; 3) considered survivors of childhood cancer by their pediatric oncologist; and 4) currently over the age of 18 years were identified and invited directly by the Janeway Pediatric Oncology Program to participant in an interview.

Data Collection Procedures

Semi-structured interview guides were developed to reflect the study objectives for both CCS and HCP by the research team (*Appendix C)*(21). The interview guides were developed and approved by the entire research team before their use. The questions were developed to address gaps identified in previous literature reviews and to meet the study objectives. Participants were given background information about the research project and primary objectives prior to the interviews. One female investigator, a doctoral student with experience in conducting qualitative interviews, completed all the in-depth interviews. Interviews were conducted in person or by telephone depending on participants' availability and location. The interviewer took fieldnotes throughout the interview. The interviews were audio recorded using the "Quicktime Player" application and were transcribed verbatim by a professional transcriptionist.

Data Analysis

We used qualitative descriptive and thematic analysis to analyze the interviews (21, 23, 24). Qualitative descriptive analysis is relevant as some of the data of interest was factual information about current transition and aftercare processes (25). For the identification of barriers to improved transition, we used thematic analysis which provided a structured method for identifying, organizing, describing and reporting themes (23, 26). In order to get familiar with all the data, we reviewed entire interview transcript before starting the coding process. We then manually coded each interview transcript. Data was initially coded by one member of the research team. The coding was then reviewed and confirmed through discussions with the other authors. After coding was complete, we organized all the data by code to identify the main themes and barriers(21). Once the main themes were identified, we then reviewed all the interview transcripts again to confirm that the themes and barriers we identified accurately represented the interview data.

Ethics Approval

Ethics approval for the project was granted by the Newfoundland and Labrador Health Research Ethics Authority (27).

Results

Of the 20 invitations sent to CCS, 5 CCS participated in an interview (Table 4.1). Interviews were conducted with 9 of the 12 HCP who were invited to participate: five adult or pediatric oncologists, three allied health professionals (dietitian, physiotherapist and social worker), and one family physician. Of the three HCPs who did not participate, two did not respond to multiple invitation requests and one was unavailable for an interview. Interviews were between 15 and 60 minutes in length. In order to maintain participant confidentiality, the demographic information of participants was not broken down further.

Table 4.1 *Participant Characteristics*

	No. (%) of Participants	
	CCS **n = 5**	**HCP** n = 9
Urban	3 (60)	9 (100)
Female	3 (60)	7 (78)
Previous Treatment		
Chemotherapy	5 (100)	NA*
Radiation	2 (40)	NA
Aftercare		
Part of Pediatric Care	5 (100)	NA
Part of Adult Care	2 (40)	NA
Medical Specialty		
Oncologist	NA	5 (56)
Primary Care Physician	NA	1 (11)
Other HCP	NA	3 (33)
Work Location	NA	
Pediatric Hospital	NA	6 (67)
Adult Hospital	NA	3 (33)

*NA defined as Not Applicable

All the CCS interviewed received pediatric aftercare until 18 years of age or older. Only two reported receiving aftercare after leaving pediatrics, with one of those only reporting being regularly screened by the cardiac program. One CCS was unsure of their specific cancer diagnosis while another struggled to recall all aspects of their treatment, which are key pieces of information survivors should know in order to help direct their future aftercare.

The Process of Transition for CCS

Through our interviews, we identified three different points at which transitions to adult care usually occur at the Janeway: 1) survivors are followed until they reach the age of 18; 2) until they are ten years off treatment (if later than the first condition); or 3) after 18 years of age but before 10 years off treatment and the survivor is deemed ready to transition by the pediatric oncologist. In all cases, decisions about transition of survivors are made in discussion with the survivor, their families and other HCPs. The transition and aftercare trajectory depend on the CCS type of cancer, treatments received and their geographic location. HCP stressed that depending on cancer diagnosis and treatment, CCS would receive specific aftercare. For example, CCS #2 received specific aftercare related to potential cardiac late effects; however, no other CCS interviewed reported other special aftercare arrangements.

Currently, there is no structured transition program in NL or regular advanced education about transition into adult aftercare. During a survivor's last clinic visit at the pediatric centre, the pediatric oncologist would usually verbally review a summary of their care with them, including their original diagnosis, treatments received, risks of late effects associated with treatment and follow-up care needed (e.g., special screening). Similarly, there are no formal transition procedures that allied health providers follow. Depending on their needs, survivors may or may not be followed by allied health providers into adult aftercare.

There are several possible destinations for CCS after they transition from the Janeway (Figure 4.2). A local family physician recently started an adult follow-up survivorship clinic (AFSC). This AFSC has been underway for approximately 3 years, with an estimated four or five CCS transitioning to the clinic annually. CCS #4 reported receiving aftercare through the AFSC. If a patient has received radiation as part of their treatment, the radiation oncologist also follows the patient alongside the AFSC. Pediatric oncologists reported that survivors who transitioned prior to the inception of the AFSC were often referred back to their family physician. Because the AFSC program is in the provincial capital city, CCS from rural communities are still usually transferred back to their family physician for aftercare. It is possible that survivors' aftercare arrangements may change over time depending on their location and other circumstances.

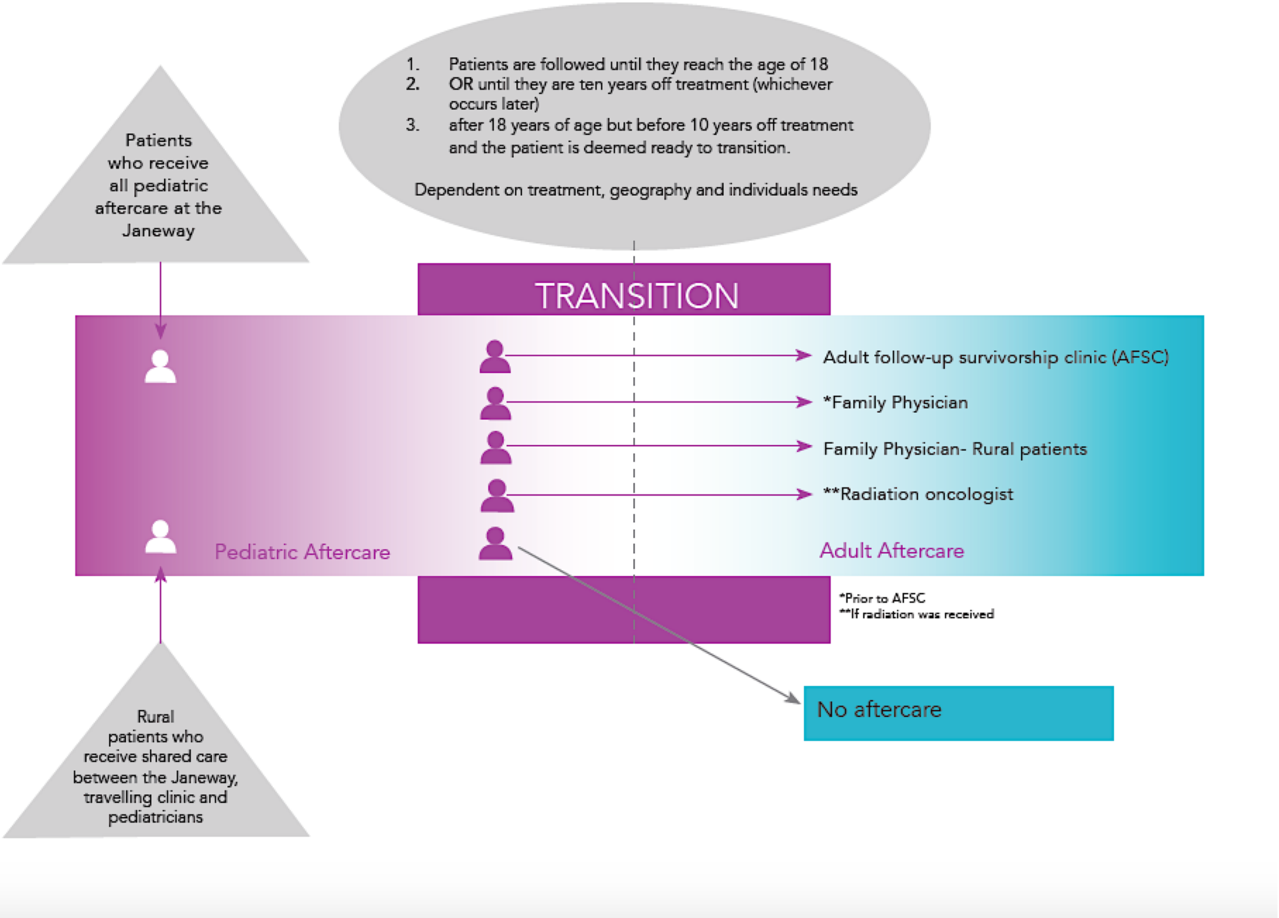

Figure 4.2 *Current Transition Process for CCS in NL**

**Original figure based on interview data*

Barriers to Improving the Transition and Aftercare

We identified the following barriers to improving CCS' aftercare experience: challenges for rural survivors, changes in availability of services after transition, challenges with navigating the adult system and lack of education surrounding transitions. *Table 4.2* highlights quotes of HCP and CCS as they relate to each theme.

Challenges for Rural Survivors

HCP and rural CCS reported considerable differences in how survivors in rural areas receive aftercare. With no pediatric care hospital outside the capital city, families reported traveling considerable distances to receive care or they receive care through travelling clinics. Survivors without a family physician in their rural community often rely on pediatric travelling clinics for all their primary care, making transition a further challenge. After transitioning out of pediatrics, participants felt there were a limited number of family physicians available to take over their care. Others reported that family doctors may not have the same understanding of cancer care as the dedicated AFSC. Survivors in rural areas also face inequities of access to allied health services and mental health support. One interviewee said "a new way of doing business" needs to be considered for how aftercare is provided in rural areas.

Changes in Availability of Services After Transition

Pediatric oncologists discussed the importance of allied health support received by CCS. Once survivors go back to their family doctor or to the AFSC, these supports are viewed as less accessible. While the adult health care system ideally has all the same services, the onus is on the patient to arrange for care and manage appointments. CCS reported having trouble accessing allied health professionals in the adult system, with long wait times to access services if they are even available. Additionally, allied health services for adults are not always accessible under public-funded health coverage programs. Without private insurance, these services can be costly and not all survivors are able to pay out-of-pocket. After leaving the pediatric care setting, CCS commonly discussed a loss of access to psychological supports. One HCP discussed the need for a multidisciplinary cancer aftercare program for adult CCS to help mitigate the loss of these services.

Challenges with Navigating the Adult System

After transitioning to adult care, CCS are expected to have a certain level of autonomy and it is up to the patient to identify their needs and arrange for appointments. From our sample, those who do not have stable, continuous housing; who do not have a regular family physician; or who still rely on their parents to organize their care often have challenges navigating the adult healthcare system. One adult provider identified some tools (i.e., patient navigators) that are provided and trained to help navigate the system for active cancer patients, although the CCS we spoke with were unaware of these resources. CCS felt they left pediatric aftercare not knowing when they would be contacted by their new provider to receive aftercare within the adult system. Despite the potential role for navigators in the adult system, previous pediatric providers are commonly still the first point of contact when issues arise in early adulthood because CCS do not know who else to contact even though they have transferred out of pediatric care.

Lack of Education Surrounding Transition

Participants said that there is currently no formal preparation for CCS transitioning in NL. The pediatric oncologist does provide a medical summary to the physician accepting the patient and provides the patient additional information including screening appointments (dependent on cancer diagnosis and treatment) and any other pertinent information related to a CCS's diagnosis or treatment. This is usually done by the pediatric oncologist who conducts the last clinic visit. HCP and CCS discussed the need for a site visit with the adult aftercare provider that the CCS is transitioning into which would occur prior to leaving pediatric care. This would include an introduction to the new HCP and additional information on how to manage their future healthcare needs. Survivors reported experiencing a general lack of communication between both the pediatric and adult healthcare systems. It was suggested that education should begin earlier in the pediatric care setting. A more formalized approach would begin before a patient's last visit and include educating the CCS about their past cancer, developing plans to help navigate the healthcare system and meeting with the receiving adult HCP while still in pediatric care settings. This approach provides the opportunity to "create a bridge to a new context." HCP recognize the need to prioritize transition; however, there is a tendency for it to get lost in amongst other health care priorities.

Table 4.2 *Key Theme and Supporting Quotes from HCP and CCS*

Key Theme	**Ident. Code**	**HCP Quote**	**Ident. Code**	**CCS Quote**
Challenges for Rural Survivors	*HCP2*	*I find that on the West Coast (of NL), or outside of St. John's, with the patients that we see in our Traveling Clinics, because, the family physicians are so in flux in rural communities that most of these patients don't have a family physician and often, we're (the pediatric medical team) the only people that they see.*	*CCS4*	*It's a big difference (being in a rural community) because it's a 12-hour drive (to the Janeway Hospital), so to come in here every year to get the check up, it was a big deal.*
Changes in Availability of Services After Transition	*HCP6*	*I wouldn't say 'loss of services'… I think that, maybe there are some services that are not as emphasized on the adult side… On the pediatric side, there's a little bit more emphasis on the sort of social work part, school, integration those sorts of things. That all of those resources are available on the adult side, I just don't think that there's as much emphasis.*	*CCS2*	*Okay, here's a doctor, here's a psychologist or here's whoever, you could go talk to, but since I haven't had that appointment (since the Janeway), I feel like I'm missing out on a lot of resources that I could… that could be beneficial to me and even when I was in the pediatric care.*
Challenges with Navigating the Adult System	*HCP2*	*I think the biggest problem for patients that are not connected to any place or person. They're not connected to a family doctor because they are moving around the province for school, or the country for school or jobs and so, they're not, they're not grounded anywhere to maintain those connections. I think if you polled most young adults, they would have no idea (how to navigate the health care system).*	*CCS3*	*(Be)cause I feel like it was more of a miscommunication in care where maybe one person thought it was being taken care of... or they thought it was somebody else's job, but I wasn't ever followed up. I was told I would have been and have something in place to transition children from the Janeway to the Health Science, or whatever the hospital that they're seen at.*
Lack of Education Surrounding Transitions.	*HCP1*	*(CCS) need to have an understanding of what their treatment was, what their diagnosis, and what are the important things to remember for their ongoing health. I think if you polled most young adults, they would have no idea. And ask them what they would do, they would all feel the same way.*	*CCS2*	*I still had a family doctor at that point but there wasn't really any contact between them. It was the whole leukemia thing was kinda dealt with in the Janeway and then like, anything outside of that was just kind of taken care of by my family doctor and he didn't really know anything about the cancer, and they didn't know anything about what was going on with my family.*

Interpretation

We examined the process of transition from pediatric to adult care for CCS and identified barriers to transition for CCS in one Canadian province for both its urban and rural populations. We found the process for transitioning had insufficient structure and lacked dedicated supports. In fact, the transition occurred as a discrete event rather than "the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult- oriented health care systems" that transition should aim to be (28-30). Only two CCS reported any form of adult aftercare. From these participants alone, we can see that there is a gap in the care that is essential to the well-being of these survivors. Other barriers to transition identified included challenges for rural survivors, changes in availability of services after transition, challenges with navigating the adult system and lack of education surrounding transitions.

Our findings reflect a situation commonly faced by CCS. A cross-Canada survey found that 88% of pediatric oncology programs reported following LTFU guidelines but only 35% had access to a formal transition program(31). Moreover, we found that CCS in rural communities face additional challenges accessing aftercare. Such inequities in rural healthcare are prevalent throughout many jurisdictions (32). Access to health care facilities influences patient outcomes (33, 34). It is also the case that direct and indirect costs associated with travel amplify the challenges for rural CCS (35). Other studies have compared CCS in rural communities with gender-matched peers and report poorer social competence, greater behavioral issues and weaker overall school performance in rural CCS (36) (37). Strategies that help reduce costs and time of travel - such a tele-health, travelling clinics and other creative services - should be further investigated to better serve CCS living in rural areas.

Education for HCP and CCS is another key feature of successful aftercare. Components of education during CCS transition that have been identified as necessary include knowledge about one's disease, disease treatment, future health risk, self-management skills and coordination of care (18, 38). Our research highlights that CCS may not have sufficient knowledge to maximize engagement with the healthcare system during transition. The Childhood Cancer Survivorship Study (CCSS) found that only 72% of CCS could correctly recall their diagnosis and only 35% were able to report awareness of any health risks (2). Ginsberg et al. reported similar findings whereby CCS were unable to report basic information regarding their health (39). Some CCS lack the skills required to advocate for themselves as they progress into adult care (40, 41). Participants in our study are supportive of an educational intervention to improve the transition from pediatric to adult care for CCS. Future research will focus on the development and evaluation of an educational intervention relevant to our study population.

Even in a province with a relatively small number of CCS, we found that providers were not fully aware of their experiences and the challenges they face after they leave pediatric care. Of the 5 CCS we interviewed, only 2 reported having received any aftercare once they entered the adult system, one of whom is only followed by the cardiac program. This is clearly not optimal nor in keeping with standards of aftercare. None of the providers we interviewed indicated that a potentially large percentage of young adults CCS in the province were not receiving aftercare.

While the current study only looked at the context for CCS in NL, it would be likely beneficial if similar work was carried out in other jurisdictions to help identify current gaps being experienced by CCS in other locations.

Limitations

While our study provides a base to make recommendations for improving care, there are some limitations. This qualitative research only studied the transition experience of survivors in one Canadian province. Caution should be used when extrapolating these findings to other jurisdictions. While we are confident in the validity of our findings, further techniques for improving the validity of qualitative research, e.g., the use of coding software and participant checking, could have also been employed. Despite best efforts and numerous methods of recruitment, we hoped that more CCS would have participated and given their perspectives. Unfortunately, we were only able to interview 5 CCS. It is not known how those who participated in the study differ in their experience from those CCS who did not. While invited to participate, neither an oncology nurse or psychologist participated in an interview, so that we could not include their perspectives on the transition to aftercare.

Conclusion

We identified the process by which CCS transition into the adult system occur in NL and identified the barriers associated with transition. Key stakeholders recognize the need to improve these gaps and offered recommendations around future interventions targeted at better supporting these CCS. Our results show that despite adherence to clinical guidelines for aftercare and high levels of satisfaction from CCS, a clear, more structured process for transition for CCS is likely needed. Our research also highlights the compounded challenges for CCS in rural communities. CCS and HCP in our study share a common goal to holistically improve the transition of CCS from pediatric to adult care. We hope that the better understanding of transition practices presented here will support the development of interventions which can more adequately prepare CCS for their transition into early adulthood and their move into the adult-focused healthcare system.

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Chapter 5

Development and Piloting of an Intervention to Improve Transition

Introduction and Overview

The goal of the WHO framework for quality improvement is to review the evidence and understand the context in order to direct the selection of an intervention for improving the quality of care (1). One of the reasons for adopting this overall approach is that it selects an intervention that is both evidence-based and appropriate for addressing the needs within a local context. In this chapter, a pilot intervention for improving the quality of CCS's experience transitioning from pediatric care into adult care in NL is presented. Prior to choosing this intervention, the research team considered its objectives, its potential impact, its ability to meet identified needs, and its development and delivery. These decisions were considered using elements of the WHO framework. The second and third parts of the cyclical process, *Strategy* and *Implementation* guided the decisions in developing and implementing the intervention. During this process, decisions were made surrounding who would need to be involved in the intervention, any potential risks, impending timelines, intervention feasibility, associated costs, and possible evaluations of the intervention.

We know that the majority of CCS who enter the Janeway oncology program will likely receive aftercare for their cancer treatments. Ideally, this aftercare will span across pediatric care and into adult care. With the addition of the AFSC, there are increased opportunities to engage and educate CCS within the local context. Many opportunities exist to intervene and engage throughout the transition from pediatric to adult care—ultimately leading to reduced late effects and improved overall health.

In our evidence reviews, overall participants felt that interventions focused on aftercare were useful and beneficial. We identified various categories of interventions. While the evidence base was not as robust as we would have hoped, interventions in each category increased patient satisfaction and were seen as valuable additions to the aftercare process. There were a number of potentially suitable interventions. Our qualitative analysis led to an education-based, transition focused intervention. Of the interventions examined, there were several that stood out. Hudson et al. completed a well-controlled educational group intervention focused on improving QOL in CCS (2-4). Their study used a randomized design and completed follow-up 3 and 6 months after the intervention. Kock et al. presented a mobile application to minimize late effects (3). Finally, Bashore et al. completed an evaluation of an interactive workbook specific to CCS transition (4). The emphasis of this intervention was knowledge surrounding diagnosis, treatment and late effects (4).

In our case study, we found that key stakeholders in NL supported the need for increased attention on transitions for CCS. Participants wanted an intervention specifically tailored towards the barriers identified in the local context. HCP and CCS who participated clearly identified barriers, including a lack of education surrounding diagnosis, treatment and risk of late effects. This finding is consistent with issues identified in our reviews of the academic literature. This consequently focused our intervention on improving education for CCS.

Transition is a complex, multifaceted process. As such, any number of interventions could have been chosen to support CCS. We ultimately focused our attention on either developing an educational workbook or a mobile application. The research team discussed the best options given the framework, timeline, resources and evidence base. Given the needs of the population, discussions with the Janeway pediatric oncology program, and the key features discussed at the beginning of this section, a workbook seemed most suitable. We also used the six domains of quality interventions as directed by the WHO framework to work through potential interventions and build a strategy on quality. Finally, we also heard from participants that online interventions might be appropriate for this population. As such, we decided to pilot a workbook and later focus on creating an electronic version of it.

In 2019, the groundwork for the workbook was started. Our workbook was informed by previous literature, other chronic disease educational workbooks, and discussions with the research team. We modelled the interactive aspect from interventions such as Bashore et al(4). For example, an entire section of the workbook is dedicated to engaging the user through activities that help develop knowledge of their diagnosis, treatment and risk of late effects. We heard from our case study participants that a local context was needed in any intervention, therefore, we used examples from other NL programs, such as a transition guide previously developed for asthma patients at the Janeway, as a guide. We also explored other transition specific resources to compliment what was already identified. The title of the intervention, *"Life After the Janeway,"* reflects a quotation heard during our case study interviews. It encapsulates the remarks of HCP and CCS that despite surviving cancer at the Janeway, care needs to continue. We believe this intervention best reflects what we heard in our situational analysis and case study, thereby maximizing the WHO strategy. We see this workbook as a realistic first step towards improving transition for CCS in NL. Additional information about the workbook can be found in *Appendix A*.

The remainder of this chapter presents an article describing our pilot workbook intervention. The article focuses on the evaluation of the intervention using the PEMAT-P instrument.

Co-authorship Statement

Evaluating a Transition Workbook for Childhood Cancer Survivors: A Pilot Study

- Devonne Ryan developed the research design supervised by Dr. Roger Chafe and Dr. Paul Moorehead.
- Devonne Ryan and Dr. Paul Moorehead recruited participants.
- Devonne Ryan drafted the manuscript.

- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead revised and critically appraised the manuscript.
- Devonne Ryan, Dr. Roger Chafe and Dr. Paul Moorehead read and approved the final manuscript.

Evaluating a Transition Workbook for Childhood Cancer Survivors: A Pilot Study

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Abstract

Background: Many childhood cancer survivors (CCS) could benefit from improved knowledge about their cancer diagnosis, the treatments received and associated risks during the period when they transition into adult aftercare. Interventions that support transition from pediatric to adult care have showed high patient satisfaction. We developed an educational workbook, *"Life After the Janeway,"* to support CCS transition into adult care.

Objective: To evaluate the understandability, actionability and overall feedback for the transition workbook "*Life After the Janeway*".

Methods: We evaluated the workbook using an online survey based on the Agency for Healthcare Research and Quality's Patient Education Materials Assessment Tool for printable material (PEMAT-P). Descriptive analysis included overall feedback, mean understandability scores, mean actionability scores and measurement of interrater reliability.

Results: Ten participants completed the survey. The overall PEMAT-P score was 94.06 (SD±7.40). Mean scores for understandability and actionability were 92.83 (SD±8.79) and 98.15 (SD±5.24) respectively. Interrater reliability found strong agreement across survey items. Participants also offered suggestions for improving the workbook.

Conclusion: Participants support efforts to improve transition and felt positively about the intervention. The workbook was shown to be understandable and actionable to likely users. Next steps will focus on delivering the workbook to CCS going through their transition of care and exploring developing the workbook in an electronic format.

Background

Developments in treatments for childhood cancer have led to considerably improved survival rates (1-3). While more children are surviving cancer, the cancer treatments they received can be damaging to the developing body and may lead to future adverse health effects (3-5). As CCS progress into adulthood, many do not continue to receive the aftercare that is recommended to monitor the late effects of their treatment (4-8). Barriers to transitioning to adult care include a shift in autonomy to the patient, challenges with navigating the adult healthcare system, lack of formal transition plan and need for enhanced education surrounding transition.

Once CCS move into adult care, they take on an increased responsibility for managing their own health. In order to do this effectively, CCS should have knowledge of their cancer, medication, treatments and potential late effects (9). Providing education supports during transition could help effectively bridge the gap between pediatric and adult care. For example, interventions to support transition have shown to be beneficial for CCS (10-13). Our research team previously completed a case study that examined the process of transition from pediatric care to adult care and the barriers to improved transitions in the province of Newfoundland and Labrador (NL), Canada (14). We found that CCS were not well aware of the specifics of their cancer diagnosis and treatment, the importance of aftercare, their risks of late-effects, or how to best manage their future care. CCS and healthcare providers (HCP) identified the need for an intervention focused on education in order to prepare for the critical transition period. Based on this research and examples found in the literature, we developed *"Life After the Janeway",* an educational workbook. In this article, our objective is to evaluate the understandability and actionability of this workbook and to acquire general feedback from participants.

Material and Methods

The Intervention

We followed the World Health Organization's (*WHO) Quality of Care: A Process for Making Strategic Choices in Health Systems* framework in the development of our intervention to support transitions for CCS (15). *"Life After the Janeway"* was developed using the step-wise process established by the WHO framework. The groundwork for this intervention began by completing systematic reviews of aftercare models and interventions for CCS. From these reviews, we found gaps in the literature with respect to transition and interventions focused on the transition period. Following these reviews, a case study was conducted to understand all aspects of transition and barriers of transition in NL. Education surrounding transition was highlighted as an area that CCS and HCP felt could be improved. After completing the case study, we focused on developing an intervention to improve education during transition for CCS based on direct feedback from interviewees. This intervention is designed to be completed by CCS and members of their circle of care during transition from pediatric to adult care.

This workbook includes three sections *(Appendix D)*. The first section is informational, and includes an introduction, description of the transition process, information on the importance of transition and what to expect from adult care. The second section is designed to be completed by the CCS and providers in their circle of care. CCS provide information related to their medical history and a list of HCP. They also complete a topics checklist, a transition readiness assessment and a health passport, which is designed to be completed in collaboration with their parents and HCP. The final section of the workbook provides health promotion advice, a list of useful organizations, a glossary of cancer terminology and a section for any notes or questions.

Research Design

The evaluation was completed using a cross-sectional survey based on the PEMAT-P *(Appendix E)*. The PEMAT-P is a tool used to evaluate two key features of educational material: understandability and actionability (16-17). Understandability is focused on patient education material being understandable to patients of diverse backgrounds and different levels of literacy (16-17). Actionability focuses on patient material being process driven for varying backgrounds and literacy levels (16-17). The PEMAT-P consists of 17 items measuring understandability and 7 items measuring actionability. The PEMAT-P provides a scale for scoring. Each question requires the participant to answer “agree,” “disagree” or “not applicable” when relevant. The statement “agree” is given a score of “1,” while “disagree” receives a score of “0.” Should “not applicable” be selected, it is not given a score, rather it is removed from the total number of questions answered. A score based on total number of questions is derived. The higher the score, the more understandable and actionable the material. Based on other research using the PEMAT-P, materials were considered highly understandable and actionable with a score of 70% or higher. This instrument has been demonstrated to be applicable to various patient education materials and useful across settings (16-18).

Setting and Participants

This study took place in the province of NL, Canada from February 25 to March 25, 2020. The survey was completed online using the Qualtrics Survey Platform (19). Participants included CCS over 18 years of age, family members of a CCS, community group/support group members, and HCP involved in cancer survivorship care.

We recruited using four approaches:

1. An oncologist, who is a member of the research team, talked with potential participants about the study during clinic appointments or follow-up. If participants were interested in taking part in the study, a recruitment letter was provided to CCS and/or parents from the oncology program.
2. Recruitment letters were provided to physicians who work within the circle of care of CCS.
3. We emailed community and support groups that CCS might be a part of and ask for their participation in our evaluation.
4. All HCP that are part of circle of care for CCS were invited to participate by email. After two weeks, a reminder email was sent to each HCP.

Procedure

Individuals who agreed to take part were provided the workbook and survey link through email. After reviewing the material carefully, participants were asked to complete an online survey. Participants were asked to review the workbook and complete the survey within one month. The booklet and survey were only available in English.

Analyses

Descriptive statistics were used to describe the sample of participants. We produced minimum, maximum, mean, and standard deviation for PEMAT-P scores. Three scores were calculated based on the PEMAT-P survey: understandability, actionability and overall. In order to calculate each score, the total sum of points for each question in the survey is divided by the total possible points (excluding the items that were scored not applicable). Interrater reliability was measured using the Fleiss Kappa statistic. Overall agreement percentages were also calculated.

This study protocol was approved by the Newfoundland and Labrador Health Research Ethics Authority (20).

Results

We had 16 participants who started the online survey. Of these participants, 10 took part in the PEMAT-P survey. One participant completed the understandability section of the survey but did not complete the actionability portion. Understandability, actionability and overall scores were calculated (*Table 5.1*). The overall PEMAT-P score and standard deviation was 94.06 ±7.40. The mean PEMAT-P score and standard deviation (SD) for understandability was 92.83 ±8.79 and actionability was 98.15 ±5.24. Interrater reliability shows the degree of agreement among raters. As such, there was strong agreement statements for understandability, actionability and overall PEMAT-P questions (85.23% and 83.33% and 83.56% respectively) (*Table 5.2*).

Table 5.1 *PEMAT-P Descriptive Analysis for Understandability, Actionability, and Overall Scores*

PEMAT-P Section	Minimum score, Maximum score	Mean Score (SD)
Understandability (n=10)	76.47, 100	92.83 (±8.79)
Actionability (n=9)	83.33, 100	98.15 (±5.24)
Overall	76.19, 100	94.06 (±7.40)

Table 5.2 *Interrater Reliability for Understandability, Actionability, and Overall Scores*

PEMAT-P Section	Fleiss Kappa (95% Confidence Interval)	Agreement (%)
Understandability (n=10)	0.78 (0.69,0.87)	85.23%
Actionability (n=9)	0.75 (0.50,1.0)	83.33%
Overall*	0.75 (0.66, 0.85)	83.56%

*data excludes participant who did not complete survey

Our results illustrate that most participants agreed with the statements laid out by the PEMAT-P survey with respect to the workbook *"Life After the Janeway."* As shown in *Table 5.3*, some participants disagreed with some statements in the PEMAT-P (12 in understandability, 1 in actionability. The most frequent disagreements were with the statement "the workbook uses visual cues (e.g., arrows, boxes, bullets, bold, larger font, highlighting) to draw attention to key points" and "the workbook presents information in a logical sequence."

Table 5.3 *PEMAT-P Statements Answered with "Disagree" and Number of Participants*

Statement	**Section**	**Number of Participants**
The purpose of the "*Life After the Janeway"* workbook is clear.	Understandability	1
The workbook does not include information or content that distracts from its purpose	Understandability	1
The workbook uses common, everyday language.	Understandability	1
Numbers appearing in the workbook are clear and easy to understand.	Understandability	1
The workbook presents information in a logical sequence.	Understandability	2
The workbook provides a summary.	Understandability	1
The workbook uses visual cues (e.g., arrows, boxes, bullets, bold, larger font, highlighting) to draw attention to key points.	Understandability	3
The workbook uses visual aids whenever they could make content more easily understood (e.g., illustration of healthy portion size).	Understandability	1
The workbook's visual aids reinforce rather than distract from the content.	Understandability	1
The workbook explains how to use the charts, graphs, tables, or diagrams to take actions.	Actionability	1

Participants who reviewed the workbook and completed the survey were asked to share their feedback and any recommendations towards improving it. Overall, the feedback was generally positive. One respondent felt it was an excellent idea and suggested that the workbook be printed in color, bound and presented as a tool for CCS and families to use for an extended period of time. Several participants felt that technology is critical for engaging adolescents and felt that incorporating the workbook into an online platform would be useful. One participant felt that the language and some practices of the workbook may be considered "too heavy" for CCS. They suggested making the workbook lighter in order to reduce fear amongst the target population. It was suggested to add a section related to survivor guilt, mental health challenges for CCS, grief management and addressing the responsibilities of the patient. One participant felt the local context of the workbook could be advanced.

Lastly, a 5-point scale was used to ask participants if they were likely to use the workbook or recommend it to others. We found that only 66% stated they were "likely" or "very likely" to use the transition workbook; however, 89% of participants would "likely" or "very likely" recommend the booklet to others.

Discussion

We created and piloted an educational workbook for CCS preparing for transition from pediatric care to adult care. Using the available literature and previous research, investigators designed this workbook to suit the identified needs of CCS in NL. It was designed to be completed by CCS with family and HCP assistance over the transition period. Overall, participants felt the workbook was understandable and actionable, suggesting that it could be a beneficial component of transition.

Of the participants who completed the survey, 89% stated that they would be "likely" or "very likely" to recommend the workbook to others, suggesting it may be a practical intervention moving forward. However, only 66% of participants said they were "likely" or "very likely" to complete the transition workbook themselves. One reason to explain the lack of potential uptake might be because the workbook was evaluated outside the context of the healthcare transition, i.e., CCS who completed the workbook had already completed their transition to adult care. Other possible reasons might include HCP who see CCS who have already transitioned and may not currently see use of the workbook but might recommend it to others.

Despite examples of interventions for CCS in the literature (10-13, 21), few of them focus specifically on transition. There is also a shortage of evaluations of these interventions. As part of a recent systematic review, we found only three examples of workbooks designed for CCS that were evaluated in the published literature (10-13, 21). Researchers of one workbook focused their attention specifically towards transition of CCS using a mixed-methods design to evaluate transition worry and readiness (21). Since workbooks are designed to deliver particular components, how well these components are portrayed should be independently evaluated. In our case, understandability and actionability were critical to the foundation of our workbook; therefore, the PEMAT-P was used to evaluate those specific components.

To the best of our knowledge, our study is the first of its kind to use PEMAT-P to evaluate an education resource for CCS transitioning from pediatric to adult care.

Participants of our study noted that some of the visual cues in the workbook could be improved to draw attention to key points. Recent literature that also used the PEMAT instrument for print and online material indicates that other healthcare studies had similar findings where participants felt visual aids were an area that could be improved (18).

This workbook is unique in the step-wise approach used to create it. We used the *WHO framework, Quality of Care: A Process for Making Strategic Choices in Health Systems,* to guide the development of an intervention (15). The WHO model provides a systematic process for decision makers in developing effective interventions for health systems (15). This model is based on a cyclical seven step process which is broken into three phases: analysis, strategy and implementation. We believe this approach helped harness the needs to CCS in the province of NL and focus on developing an intervention designed with users, for users.

Participants recommended the need for an electronic version of the interventions. Previous work has found there is an increased demand for these types of interventions in the medical field. The rapid development of technologies available to youth and adolescent provides an opportunity for ease and accessibility. Several platforms are possible including a mobile application. While evidence of the efficacy is limited with respect to mobile health applications, it offers an easily accessible medium for adolescents. While an online platform for this type intervention may seem innocuous, there are challenges to be overcome, e.g., secure storage of patients' private medical information, secure networks and universal platforms. Identifying and managing these challenges would be an important part of future research in this area.

There were several strengths to our study. Our workbook is the first of its kind for CCS in NL. We used a well-established instrument, the PEMAT-P, as a tool for evaluation. We also used an established framework, the WHO *Quality of Care: A Process for Making Strategic Choices in Health Systems* (15), to guide the creation of the workbook.

There were several limitations to this study. Given the small population of HCP who care for CCS and the small population of CCS in the province, we chose to try to reach participants through convenience sampling. Despite best recruitment efforts, our sample size was small. Although our sample size is small, we believe it is adequate given the nature of our research and our results. Overall PEMAT-P scores were considered high with no value below the cutoff of 70. The mean score of 94.06 with a standard deviation of 7.40 suggests that scores remained above the cut-off value. We believe the information gathered is important for continuing the implementation and monitoring elements of the WHO framework. Other research similar to this workbook by Bashore and Bender (2016), which had a sample of 30 participants, reported only 20 who completed the study (21). The Centers for Disease Control (CDC) used PEMAT-P and a sample size of 10 to assess their "Sepsis Patient Education Material. (18)." Results were consistent between participants; therefore, an expanded sample size may not have had any benefit. Several participants started our survey but did not complete it. Unfortunately, this is one disadvantage of online surveys and not unique to our study (22). This was a single-institution pilot study and the findings cannot be generalized to other centres. We plan to implement the workbook in a clinical setting and modify it to allow for evaluation in other healthcare centers. Finally, we did not study CCS who are currently experiencing a transition from pediatric care to adult care. Future steps of this research will focus on these users.

Overall, the "*Life After the Janeway"* workbook received positive feedback from the survivorship community. The findings of this pilot highlight that the workbook is understandable, useable and may be a beneficial component of a formalized transition program. We plan to generate an updated version that incorporates participant feedback. This version will include an added section addressing survivor guilt, mental health challenges, grief management and the responsibility of the patient. We will revisit each section to confirm it is tailored to a local context. Finally, we will explore our ability to create an online version and mobile application of the workbook. Following these changes, we intend to complete a RCT that evaluates the effectiveness of the workbook using pre-defined education-based outcome measures. This study will focus specifically on CCS using a pre-posttest design to measure outcomes as CCS transition from pediatric care into adult care. We intend to overcome our limited sample size by implementing this study in other institutions across Atlantic Canada. In order to maintain the local context, we will work with key stakeholders in other jurisdictions to ensure the workbook meets the needs of those populations.

Conclusions

It is critical that CCS transition from pediatric care to adult care with knowledge of their disease and potential late effects of their treatment. Poor transition can lead to morbidities and poorer health outcomes later in life. Education for this population is an important practice to help reduce the burden to CCS long-term. This study shows that participants found the workbook *"Life After the Janeway"* to be understandable and actionable. We believe this workbook is a step in the right direction for improving long-term care and management of potential effects in CCS, which could be adjusted and adopted by other cancer programs.

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Chapter 6

General Discussion and Implications for Practice, Policy and Research

Overview and Main Findings

There is a scarcity of academic literature exploring transitional practices for CCS, specifically in NL. This thesis project concentrates on developing an in-depth understanding of aftercare practices and interventions; understanding how transition is currently being carried out in NL; identifying barriers for transition in NL; and completing a pilot intervention to promote better care at this critical time in survivors' lives (1-3). In order to meet these objectives, two systematic reviews were completed (4, 5) (found in Chapter 3), an extensive qualitative study (found in Chapter 4) and an intervention tailored to meet the specific needs of the NL population (found in Chapter 5). The approach was guided by the WHO "*Quality of care: a process for making strategic choices in health systems(6)"* framework. This chapter includes a discussion of the main findings of each phase of the research, the potential policy implications of this work, the experience of using the WHO framework, knowledge dissemination, and the strengths and limitations of this research.

The objectives for this thesis project were 1) to review MOC and interventions in the literature; 2) to provide a detailed description of the process of transition in NL; 3) to understand the experience of transition in NL; 4) to identify limitations, barriers and gaps in services and practices in NL; 5) to pilot an intervention aimed at improving transition; and 6) to disseminate our findings. All these outlined objectives were met, and a comprehensive understanding of the transition from the Janeway's pediatric oncology program into adult care in NL was achieved. These findings highlight the complexity of transitions and the need to support CCS and HCP during this period in a patient's journey.

Categorization Paper and Systematic Reviews

Many critical components of aftercare were found. With increased demand for these aftercare models and inconsistencies in how they have been categorized before, the need to develop a model to help categorize important aspects of MOC was determined. The article, *"Standardizing the Categorizations of Models of Aftercare for Survivors of Childhood Cancer,"* focused on developing a way for researchers studying MOC to evaluate components of each model in a consistent approach. This article presented a novel taxonomy to classify and describe models based on six key aspects we found in the literature: lead provider, other providers involved in delivering aftercare, where care is provided, how are survivors engaged, which services are provided, and who receives aftercare.

The systematic review, entitled *"Models of care for childhood cancer survivors once they become adults: A Systematic Review,"* grouped MOC used by CCS and examined the evidence supporting each model (4). The review included 22 articles. Data was extracted from each article including models of care / program described or evaluated, geographic location, number of participants, type of study participants, key features of MOC studied, outcomes evaluated and conclusions of each included article. The primary MOCs inferred from our analysis included pediatric-led, adult oncology-led, PCP-led, hybrid oncology/primary care, nurse-led and other-led follow-up. This review supported the importance and need for MOC for CCS; however, the evidence was mostly limited to patient and HCP satisfaction. This review determined that MOC were specific to location and populations of CCS, providing more reason to study the NL population closer.

The review, entitled *"Interventions to improve the aftercare of survivors of childhood cancer: A Systematic Review,"* summarizes the evidence of the effectiveness of once off interventions to improve aftercare for CCS (5). Twenty-nine articles met our outlined inclusion criteria. These articles were grouped together based on the type of intervention. These groups included social skills development, physical activity, workbooks, education and web-based interventions. Interventions were ranked high in terms of patients and provider satisfaction; however, there were clear gaps in the literature, including a lack of interventions to support transition.

Overall, across the reviews, the transition from pediatric to adult care in CCS was not sufficiently studied. Following the WHO framework, a qualitative study was designed to better understand transition from pediatric to adult care in NL.

Qualitative Study

Using a case study design, the transition from pediatric aftercare to adult aftercare for CCS (7) within its real-world context in NL was investigated (8). This case study, entitled *"Transition and Aftercare for Survivors of Pediatric Cancer across a Canadian Province,"* is the first of its kind to focus on uncovering the multi-layers of transition for CCS in NL. The case study included 14 interviews with HCP and CCS. The primary objectives were to develop a holistic understanding of how transition occurs and to understand the barriers of transition for CCS from the perspective both of CCS and HCP. Four main barriers to improvements in care in NL were identified: 1) added challenges for patients in rural areas, 2) changes in the services available for transition, 3) challenges with navigating the adult system, and 4) lack of education surrounding transition and aftercare. It was apparent from this case study that the transition from the Janeway oncology program to adult care could be improved. Participants voiced their support of improving transitions through developing potential interventions. Using the results of the case study and evidence of support stemming from our systematic reviews, an intervention was developed to address one of the outlined limitations: the lack of educational materials.

Intervention

Based on the results of the case study and the guidance of the WHO framework, an educational intervention in the form of a workbook entitled *"Life after the Janeway."* was identified and developed. The workbook focuses on three areas for CCS:

information, management and *resources*. The workbook provides important information about transitions, managing late effects and provides interactive management skills including a medical record, transition readiness assessment and health recap. As this is a pilot project, the PEMAT-P tool was used to evaluate the usability and understandability of the intervention. The evaluation was completed by CCS, their families, community groups and HCP. Mean scores for understandability and actionability were 92.83 (SD±8.79) and 98.15 (CI±5.24) respectively. The overall PEMAT-P score was 94.06 (±7.40). Interrater reliability found strong agreement across survey items. These PEMAT-P results highlight strong understandability and actionability in the workbook. Despite some critiques and feedback, participants supported the need to improve transition and were satisfied with the intervention *"Life After the Janeway."*

Strengths of this Research

Flexible methods were adopted allowing us to best understand the complexities in the delivery of transition services to CCS. Following the WHO *"Quality of care: a process for making strategic choices in health systems (6)"* framework was critical for guiding this research. This previously established framework provided a semi-structured outline which allowed for flexibility across methods for the local context. Similarly, using various approaches and increasing the number of research strategies used throughout our project, allowed the scope and dimensions of the research to deepen (9, 10). Using multiple methods, including systematic reviews, case study design, and survey design provided a richer evidence-base to support the development of an educational intervention tailored to this specific population (11). This research adhered to guidelines set forth by the Health Research Ethics Authority (refer to *Appendix F* for additional ethics documentation). In the rest of this section, the strengths of each stage of the research will be described (for more detail, please refer to Chapters 3, 4 and 5).

There were several strengths of our systematic reviews. These strengths are outlined in detail in each of the manuscripts found in Chapter 3. Importantly, the team included researchers with expertise in the systematic review process, including a librarian who helped build a strong search strategy. The study was built around a clearly defined research question and focused inclusion and exclusion criteria. Additionally, the PRISMA guidelines for systematic reviews were followed and transparency was maintained in in each step of the process. Finally, the study took several precautions to overcome bias. Multiple reviewers were used during the study selection process and throughout the synthesis of results. Moreover, multiple reviewers were used at each stage of the research to determine study inclusion, to assess the quality the studies, to complete data abstraction and to synthesize the data. Finally, the results were robustly synthesized to provide a deeper understanding of the current academic literature.

The case study also had several strengths. First, a holistic understanding of how transition occurs in NL was captured, mainly because many key stakeholders, including HCP and CCS were reached. Both rural and urban populations in the province were captured. Additionally, a substantial amount of time was spent developing an open-ended question structure that allowed for us to adapt to differences and diversity across participants. Trustworthiness was established in this study by ensuring credibility, transferability, dependability, confirmability, and authenticity (12). Credibility was established by the use of standard qualitative methods. This was further validated by effectively outlining a noble study rationale. Aspects of this study may be applied to other settings or chronic diseases; as such, presenting the element of transferability. Dependability was founded through the use of a variety of HCP interviews which confirmed the reliability of the data. Confirmability was recognised through the consistency of findings across interviews. Lastly, authenticity was established by including a representative sample of participants across the continuum of aftercare. This included participants within the pediatric and adult health care systems.

The intervention, *"Life After the Janeway,"* was developed using information garnered from the targeted population. The workbook addresses a key concern identified by HCP and CCS in NL; therefore, the results were used to develop a location specific intervention to improve transition. The intervention also follows the guidelines set forth by the CPS and American College of Physicians as discussed in Chapter 2 (13-15). Self-efficacy was identified as a major component of appropriate self-management in healthcare. The intervention focused on accountability for components of past health and future needs (16). Patient and HCP relationships at the Janeway were important influences of CCS experience. The pilot intervention is an example of a practical application to support this population and has the potential to address key concerns surrounding education of diagnosis, treatments, and the importance of aftercare. The assessment tool, the PEMAT-P, is a previously established and validated instrument to assess understandability and actionability. Previous studies have assessed the tool and showed that it demonstrates strong internal consistency, reliability, and evidence of construct validity. One of the greatest strengths of this research is its practical focus in turning evidence into a solution to address a need identified by patients and key stakeholders.

Research is a collaborative and team-based process and collaboration in health research is a valued mechanism for strengthening research and building knowledge. DR was the principal researcher for this dissertation. Each manuscript however outlines the various roles of team members.

Limitations of this Research

Several limitations have been identified, briefly described below. (for more details, please refer to Chapters 3, 4 and 5).

There were several limitations to our systematic reviews. The intention when we set the inclusion and exclusion criteria was to allow for variation in the research designs by including both quantitative and qualitative studies. It was felt that allowing for a diverse range of study designs would help uncover the current field of literature and maximize the findings. While these reviews did not fit the traditional systematic review design, the diverse forms of evidence offered increased relevance. Unfortunately, one of the drawbacks to this type of research was an inability to collate and synthesize results, due to the variety and heterogeneity of the evidence. Other researchers have discussed the challenges and benefits of these types of reviews (17-19). Other issues included the broadness of our search strategy. This led to the exclusion of many articles that were not relevant. The review was restricted to peer-reviewed, published studies, and we did not include grey literature in this field, running the risk of publication bias. The review was also limited to articles published in English and to full articles indexed in the identified article databases. Heterogeneity was also an issue across studies, making it challenging to compare and contrast the studies included in our review. Quality assessments and tables which included key characteristics for each study were completed. Using these tables, the similarities and differences across studies were outlined. However, there is no doubt that search restrictions increased the risk of missing potentially relevant articles and reduced the completeness of the review.

This qualitative study was subject to several limitations. As with any single case study design, there were issues pertaining to methodological rigour that are debated throughout the literature. Yin discusses one of the biggest criticisms of case study research is the absence of systematic procedures and methodological guidelines (7). Using the WHO framework in tandem with the case study design helped maintain rigidity in our processes. Another limitation of the case study includes issues around construct validity. Construct validity is the evidence that a test is measuring the construct of which it claims to be measuring (7). Ideally, we would have used other methods of data collection to compare data. Despite this, we did find that a diverse group of participants contributed comparable and similar data. There were challenges recruiting CCS for this study. After limited success in recruiting by mail, the pediatric cancer clinic directly contacted CCS to participate. Given our small sample of CCS, it is not likely that data saturation was reached with his population. It is possible that those who chose to participate in our study were more motivated than those who did not take part. Therefore, intrinsically, this population may have different characteristics than CCS who are not represented. We also recognize that there are challenges with respect to reliability and replicability in a single case study analysis. There may also be concerns of external validity or generalizability. The results presented are specific to the population of NL. Despite this, reporting these findings offers an understanding into different ways that care is carried out for CCS.

The success of the intervention was pleasing, despite some noted limitations. The goal was to pilot an intervention and garner general feedback and information on the understandability and actionability of the workbook. Given the small population of CCS and HCP serving this population, detailed demographic information could not be collected due to privacy concerns, therefore the data could not be analyzed by subpopulation. The evaluation survey was cross-sectional in nature as we used the PEMAT-P tool. Similar to other phases of this research, there was a potential for selection bias. The population who agreed to take part in the intervention may have differed from those who did not. This study is also limited in its small sample size; as such, the results of this study should be interpretated with caution. Lastly, this intervention is currently available only in a printed form. With information constantly changing, it is challenging to keep a printable document up to date. In the next stages of this research, it is hoped that online or electronic materials will become available to ensure that the intervention can be constantly updated.

Another challenge was the open-ended nature of the research field. Childhood cancer survivorship is an area of research with abundant possibilities and no clearly defined end point. To help mitigate this challenge, an established framework was followed, various levels of research were diligently completed, and robust effective clear collaboration with the research team occurred seamlessly.

Experience with WHO Framework

The WHO's *"Quality of care: a process for making strategic choices in health systems" framework* was chosen because it allowed for flexibility within its methods (6). The framework aligned well with the study objectives. A main reason for choosing this framework was the strategic responsibility component, which gives decision makers a process for developing and implementing interventions. This framework followed three key steps for building a strategy for quality improvement: analysis, strategy, and implementation (6). These steps aligned with the goals of this research and each step helped build capacity for the next steps. By breaking down each component of the framework, a gap in care was identified, and the research focus was then on improving that area in subsequent steps. Under the analysis stage of the framework, the involvement of all key stakeholders as suggested by the WHO was critical for the success of this project (6). The strategy stage of the framework provided a solid foundation of important pieces for the intervention. The domains outlined by the WHO framework were paramount in ensuring the intervention focused on all aspects that were important to our target population. Finally, when it came to the implementation stage the WHO provided a clear outline how to structure the implementation and monitor the process for the intervention. The WHO framework was well defined and easy to follow. No drawbacks were encountered following this framework. As such, we would encourage other researchers to use this framework when developing interventions.

Our Findings and Current Literature

The benefits of aftercare and transitions for the CCS population have been well documented. Recommendations for carrying out aftercare for CCS is evolving, but practice is not yet well established in all jurisdictions (14, 15, 20, 21). Similarly, research on the effectiveness of interventions remains an understudied area and there is a gap between research and practice in public health (22, 23). In this section research that exists in this field will be discussed, areas that may be improved on will be described and the ways this research helps address some of the gaps in the current literature will be addressed.

The research started by completing two systematic reviews to holistically understand aftercare. After completing the systematic reviews, we found that although there was a plethora of literature related to models of care, interventions and risk of late effects for CCS, there was a shortage of high-quality literature related specifically to the transition between pediatric and adult care. Similarly, we found there was a lack of a standardized basis for categorizing CCS aftercare. By developing a standardizing method of categorization in the field, (see Chapter 3), this research has provided the means for more focused direction to future assessment of this topic.

The results of our systematic reviews and the extensive assessment of the literature were used to build a case study to examine the process and barriers of transitions in NL. To the best of our knowledge, our case study and intervention are the first of their kind for CCS in NL. However, the results mirrored issues that were found in the literature surrounding transitions. In particular, researchers have clearly identified that CCS demonstrate a lack of knowledge related to their diagnosis, treatment and late effects (24-26). Through interviews with CCS and HCP, this was found to also be the case at the Janeway. Moreover, our case study highlighted the need for more information surrounding future risks which are also well supported within the literature (27-29). While many of the issues surrounding transitions aligned with previous research, we did find CCS and HCP had NL specific issues including challenges with rural care and issues with navigating the healthcare system.

In developing our intervention, we used as guides the existing workbooks alongside the findings from review articles, the case study and the WHO framework. This approach is novel in its step-wise approach. Examples of transition programs throughout Canada were found in the literature. While these programs were not cancer specific, they offered valuable insight into the importance of focusing on transition. One example, ON TRAC, focused on self-advocacy, self-esteem, independence, social support, education, planning and life style behaviors' (30). Similarly, the Hospital for Sick Children in Toronto, developed a program for adolescents with chronic conditions transferring from pediatric to adult care. They focused on development, leadership and changing foci (30). These programs exemplified the value in planning for transition and offered an opportunity to engage adolescents and promote health behaviors, in turn, minimizing health burdens in the future. In other literature, an emphasis was placed on planning for transition long before a patients last visit (14, 31). There are several examples of workbooks and material that were used to develop our intervention. Some examples of the materials that were used to guide the creation of our workbook include the Janeway's "*Asthma Transition Guide,*" Markham Stouffville Hospital's *"Young Adult Transition Guide"* for transitioning diabetes patients (32), Sick Kids Toronto *"Good to Go"* tools and resource material (33) and *"Got Transition"* material (34). Many of the workbooks that were available were not specific to transitioning for CCS; therefore, our workbook combined features of previous material while tailoring it to suit the needs of CCS in NL. The research contained in this thesis exemplifies the importance of preparation and education for adolescents as they transition and offers a unique approach to support CCS in NL, while making a new contribution to the educational resources that are available to young adults during their transition into adult care.

Policy and Clinical Implications

This section addresses potential implications that this research has on clinical and policy practice. Knowledge in the field of childhood cancer and survivorship continues to improve as medicine advances. With this, evidence-based guidelines and interventions are evolving. Appropriate interventions and guidelines, such as the COG, CPA and the American Pediatric society, should be applied in unison (14, 15, 20). Formalized transition models should be developed to help CCS into the adult health world and reduce the risk of losing them in adult aftercare (35). Findings from the studies in this dissertation have practical implications that serve to enhance the provision of transition from pediatric to adult care and hopefully overall clinical management of CCS into adult aftercare programs. For example, using the evidence garnered from our reviews and case study, it is hoped that the workbook can be implemented into practice to educate CCS early and provide them an avenue to engage in their healthcare goals. Our hope is that by promoting education around transition early, CCS are able to better manage their healthcare practices into adulthood and in turn, reduce their risk of late effects not being identified and treated appropriately.

Transition of CCS into appropriate adult care has implications on a health policy level. Ultimately, CCS should receive coordinated and comprehensive health services that go beyond the current level of care in NL. At a minimum, all CCS should receive some form of confirmative follow-up. The intervention in this thesis is only one piece of continued engagement with CCS. The intention is to build on the outlined challenges of transition. Formal healthcare supports should be dedicated to CCS at this time in order to ensure that health services are individualized and appropriate. Interventions that support the transition to aftercare, such as "*Life After the Janeway",* should be further explored and importantly evaluated. The results of the intervention in this thesis should be expanded past a pilot and applied to future CCS populations. Information in this field is evolving and an opportunity to expand on this intervention exists. There were several other gaps in transitions for CCS identified in our qualitative research. These included challenges with navigating the healthcare system and challenges for rural CCS. Looking towards the future, researchers face several challenges in advancing the field of survivorship research. The final challenges that researchers and stakeholders are faced with is how to best disseminate and use the information garnered from research in this area.

Research Implications

The findings presented in this dissertation have implications for future research in the field. As survival rates for CCS are substantially greater than 20 years ago, research should continue to monitor CCS for the long-term effects of cancer treatments. Examination of the impact of successful transition should be considered alongside the effects of monitoring for late effects. Transitions remain a grossly understudied area and should be examined closely at each level to improve the direction of health for CCS. Given the importance of adhering to guidelines and recommendations, it is imperative to continue to monitor how HCP and CCS interact with the health system during transitions. Future research should consider pursuing improved coordination of care during the time of transition and monitoring the timing at which initial contact and conversations occurs between CCS, their families and HCP.

These research findings unveiled other barriers outside of education that should be addressed. Further, the research offered insights to the challenges that CCS from rural communities' face. Opportunities to enhance the care of CCS in rural communities of NL should be explored. Qualitative information gleaned from the case study data can then be used to support additional studies to address barriers of transition for CCS. These challenges were seconded by CCS, where they offered suggestions to help mitigate these issues. Opportunities to use technologies such as tele-health to provide follow-up should be researched.

As new generations of CCS transition to adult care, it is essential that research findings support their information needs. Providing information to a new generation of cancer survivors might mean developing online platforms to engage adolescents. Educational resources using mobile applications could be used and evaluated. We heard from CCS and HCP that incorporating mobile applications and online supports could be useful to support CCS and their families during transitions. After completing the pilot, it is hoped that this research can be expanded to explore online platforms.

The intervention, *"Life After the Janeway,"* should continue beyond the pilot stage. CCS and HCP valued the workbook and results from the PEMAT highlight its understandability and actionability. Next stages of development should include an examination of the interventions influence on improving measurable transition outcomes. Outcome measures to identify important aspects of an individual's knowledge of disease, treatment of disease, knowledge of late effects and transition readiness should be determined. A rigorous study design should be used, for example, a waitlist control trial or a randomized control trial. Development and evaluation of transition programs and interventions offer researchers an opportunity to study long-term effects of education during transition on long-term health across the entire lifespan. Research should be evaluated and strategies should continue to evolve in an effort to maximize QOL for CCS.

Knowledge Translation

This research yielded valuable contributions to the field of cancer survivorship. Knowledge exchange, synthesis and application of findings is critical to ensure key stakeholders in the cancer field have the opportunity to engage with and learn from our research. Knowledge transition of this research is focused on three key audiences. CCS and their families are at the forefront of the dissemination. The focus should also be on HCP and policy makers. This research will be presented to these key audiences in order to disseminate the results and determine next steps. An invitational workshop is planned to discuss our findings and explore ways that they could be put into practice within the current healthcare environment.

As an initial step to facilitate improvement of the transition from pediatric aftercare to adult aftercare in NL the results of this thesis have been published and submitted to peer-reviewed journals (See publication details in Chapters 3, 4 and 5). Further, we expect to disseminate our findings by attending conferences relevant to this field including the *Cancer Survivorship Conference* and the *Canadian Centre for Applied Research in Cancer Control Conference.* Collaborations and partnerships will be created with key stakeholders including CCS, HCP, policy makers, and government agencies. Creating such partnerships allows various levels of care to connect and create open lines of communication in order to best support the NL population of CCS and maximize the continuity of care, effectively strengthening the healthcare system.

The impacts of poorly managed aftercare and late effects can be debilitating for CCS. Knowledge translation is a complex series of interactions between researchers and stakeholders which we hope to continue through interactive processes. Similarly, we believe that continued communication, knowledge utilization and synthesis between parties can allow for continued implementation and development of our intervention. The connections that have been made through the research team will help to promote implementation and knowledge translation. Through quality knowledge translation processes, we hope to influence forthcoming research in the field.

Conclusions

Despite surviving cancer, CCS continue to be at risk of late effects of their treatment. Ensuring adequate processes and education are in place may mitigate these risks. Therefore, health promotion practices should be supported to improve transitions and aftercare. Being proactive and promoting early transition preparation can help overcome some of the challenges faced by CCS. We were able to bring light to these issues through our systematic reviews and highlight the barriers specifically in NL using the case study method. Finally, our intervention focused on overcoming barriers of transition for CCS and their families through developing an intervention aimed at improving knowledge around their diagnosis, treatment and aftercare needs in order to optimize quality of life.

The findings from this study are valuable because they demonstrate a lack of process and focus on transitions for CCS in NL. CCS and HCP identified specific barriers to care during transitions, not all of which were addressed through our intervention. An opportunity exists to enhance care by addressing other barriers outlined in this dissertation. Although results of this study are specific to CCS, challenges faced by adolescents during their transition from pediatric care to adult care may be relevant for adolescents with other chronic conditions.

The far-reaching implications of a poor transition are well documented in the literature and place CCS at risk of wide spanning risks including delayed diagnosis and treatment for late effects (36, 37). Childhood cancer survivorship is a lifelong condition that requires education, prevention and monitoring throughout the course of CCS lives. Pediatric and adult HCP in this study shared a common goal of providing CCS with optimal transition practices. Moreover, they discussed the value of preparing CSS to take control over their healthcare and independently be able to manage their aftercare. This research, including the case study narratives and intervention, serves as the foundation for the journey to improve transitions for CCS. Future research should continue to engage CCS through interventions that target education and other barriers for transitions to optimize aftercare practices.

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Appendices

Appendix A

Additional Information for Methodology of Studies

This Appendix provides further methodological details about studies that have been published; it was not possible to include this information in the published manuscripts.

Chapter 3: Models of care for childhood cancer survivors once they become adults: A Systematic Review

The full review protocol, was reviewed and approved by a university librarian, two experts in conducting systematic reviews, and the full study team before being initiated.

Eligibility Criteria

Articles were included if they met the following criteria:

- Were articles about the health services and programs of care provided to survivors of childhood cancer, including transition programs, monitoring programs, integration of care, and models of care;

- Were articles focused on survivors of childhood cancer
- Presented original empirical data (e.g., survey / interviews) from survivors, family, healthcare providers, or other people;
- Were published between January 1, 1995 – September 13, 2017;
- Were published in English

Articles were excluded if they met the following criteria:

- Focused on evaluations of specific clinical interventions; accounts of patient need; diagnosis / assessment tools of symptoms;
- Were not published articles (e.g., exclude abstracts, review articles) *

Search Strategy

After establishing the inclusion criteria, two researches met with Health Sciences librarian at Memorial University of Newfoundland who assisted with the search strategy. The final search strategy was as follows:

((child*[tw] OR adolescent*[tw] OR pediatric[tw] OR paediatric[tw]) AND (survivor*[tw] OR aftercare[tw] OR "long-term follow-up"[tw]) AND (cancer*[tw] OR malignan*[tw] OR Neoplasms[Mesh]) AND (program*[tiab] OR service*[tiab] OR support*[tiab] OR "delivery of care"[tw] OR "model of care"[tw] OR "models of care"[tw]) AND (1995:2017[dp]) AND (English[lang]))

We performed a structured literature review of the PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo. All database searches were completed and reviewed with the assistance of the librarian. The importing of abstracts and references into the RefWorks was also completed by Devonne Ryan with the assistance of the librarian.

Study Selection

For this review, we focused on models and programs of care, rather than one off or short-term interventions to improve the survivorship experience, which are covered in a subsequent article. Searches were conducted between June 26, 2015 and September 13, 2017. We removed all duplicate titles using the delete duplicates function in RefWorks. All titles were then initially reviewed by two researchers (Devonne Ryan (DR) & Yoshani De Silva (YS)) to remove irrelevant articles. Any questions about whether to include a title in the abstract review were made by consensus between the two reviewers. All potentially relevant abstracts were then reviewed by three reviewers (RC, DR, & Paul Moorehead (PM)) with decisions about their inclusion made by consensus. The full articles for all included abstracts were then reviewed by two reviewers (RC & DR). The reference lists of all included articles were also reviewed to identify further articles which may have been relevant to the review, of these references one was included in the final review.

Data Extraction

Data extraction was completed by DR and RC. This data was used to illustrate the results of study. The data extraction table is presented in Table 3.3. Additional data was extracted by Devonne Ryan and Roger Chafe, including a table that categorize MOC (Appendix A). The information in this table includes:

- Which provider is primarily responsible for aftercare
- Which providers are regularly involved in providing aftercare
- Where is care provided
- How are survivors engaged
- Which services are provided
- Who receives services

Quality Assessment

Because of the type of studies identified in the review, we used the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) to assess the quality of each article. There are several study designs found in observational research. STROBE's aim was to create a checklist of items that should be included in articles reporting such research. As such, we created a table with the 22 items in the STROBE checklist and assessed each of our included articles. Overall, we found the methodological quality to be low. Most studies were cross sectional and single site studies. Some of the included studies had large sample sizes and followed a rigorous study design. Almost all of the included studies had clearly defined title, abstract, methods. Despite this, several of the studies lacked descriptive data, information on participants, and outcome data in the results section.

Chapter 3: Interventions to Improve the Aftercare of Survivors of Childhood Cancer: A Systematic Review

The full review protocol was reviewed and approved by a university librarian, two experts in conducting systematic reviews, and the full study team before being initiated.

Eligibility Criteria

We performed a structured literature review of PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo databases.

Studies were included if they:

- Described or evaluated a psychosocial, educational, transition, physical activity, or health behavior modification intervention provided to CCS
- Presented original empirical research;
- Were published between January 1, 1995 and September 13, 2017; and
- Were full articles, published in English.
- Focused on survivors of childhood cancer
- Presented original empirical data (e.g., survey / interviews) from survivors, family, healthcare providers, or other people;
- Were published between January 1, 1995 – September 13, 2017;
- Included only published articles (e.g., exclude abstracts)

- Were articles in English

Studies were excluded if they were:

- Evaluations of clinical interventions provided as part of clinical care
- Evaluations of models of care or programs of care
- Accounts of patient need
- Diagnostic or assessment tools
- Not published in English
- Published outside of set dates (June 26, 2015 and September 13, 2017)

Search Strategy

After establishing the inclusion criteria, two researches met with Health Sciences librarian at Memorial University of Newfoundland who assisted with the search strategy. The search strategy was reviewed and approved by a university librarian, two experts in conducting systematic reviews, and the study team before being initiated. The final search strategy was as follows:

((child*[tw] OR adolescent*[tw] OR pediatric[tw] OR paediatric[tw]) AND (survivor*[tw] OR aftercare[tw] OR "long-term follow-up"[tw]) AND (cancer*[tw] OR malignan*[tw] OR Neoplasms[Mesh]) AND (psychosocial*[tiab] OR education*[tiab] OR transition*[tiab] OR physical activity tw] OR behavior modification[tw]) AND (program*[tiab] OR service*[tiab] OR support*[tiab] OR "delivery of care"[tw] OR "intervention"[tw] OR "interventions"[tw]) AND (1995:2017[dp]) AND (English[lang]))

All database searches were completed and reviewed with the assistance of the librarian. We performed a structured literature review of the PubMed, EMBASE, CINHAL, ERIC, and PsychoInfo. For this review, we focused on interventions, rather than models of care to improve the survivorship experience, which are covered in a subsequent article. Searches were conducted between June 26, 2015 and September 13, 2017.

Study Selection

All titles were then initially reviewed by two researchers (DR & YD) to remove irrelevant articles. Any questions about whether to include a title in the abstract review were made by consensus between the two reviewers. All potentially relevant abstracts were then reviewed by three reviewers (RC, DR, & PM) with decisions about their inclusion made by consensus. The full articles for all included abstracts were then reviewed by two reviewers (RC & DR). The reference lists of all included articles were also reviewed to identify further articles which may have been relevant to the review, of these references one was included in the final review.

Data Extraction

The research team developed a standard data extraction form using the outlined study objectives. The following items were extracted from each study:

- Author, year location
- Type of intervention
- Intervention summary
- Study type
- Participants
- Outcome measures
- Conclusions from article

Quality Assessment

To assess the quality of each article we used Strengthening the Reporting of Observational Studies in Epidemiology (STROBE). STROBE's aim was to create a checklist of items that should be included in articles reporting such research. As such, we created a table with the 22 items in the STROBE checklist and assessed each of our included articles. Many of the articles included in this study used a randomized study design. Despite this, there were some issues related to quality of studies. These included low sample sizes, lack of clearly defined outcome measures, and minimal follow up periods. Additionally, many of the articles were pilot studies of interventions. For the studies included in this article, overall, a summary of the main results were well interpreted in the discussion.

Chapter 4: Transition and Aftercare for Survivors of Pediatric Cancer in Newfoundland and Labrador: A Qualitative Study

Study Design

We used a qualitative research design, specifically, a Case Study design for this research. The case study is an established research design that is used extensively throughout a variety of fields. A case study design can be defined by "the need to explore an event or phenomenon in-depth and in its natural context. It is for this reason it is sometimes referred to as a "naturalistic" design (1) ." In the case study design, it is common that participants provide in-depth and multi-faulted insight into aspects of the case under examination. Yin describes case studies as a means to explain, describe or explore events or phenomena in an everyday context (2). This can be used to better understand and explain such events or phenomena. The case study is focused on the "how," "what," and "whys" of research. These questions align succinctly with our research objectives: *How can we understand transition in NL? What are the described barriers to transition from the perspective of health care providers and childhood cancer survivors?* Further, the case study offers insights into potential gaps within an event. These aligned well with our objective of identifying barriers to transition for CCS. We conducted key informant interviews of CCS and HCP. Our chosen approach allowed for a complex and deeper understanding of the issues. Further, we chose this approach to showcase a report that included the voice of the participants, the reflexivity of the researchers, a description of the process for transition, and the barriers of transition.

Defining the Case

Our research objectives, previous research, and existing literature were considered when concisely defining "the case." The case we chose to examine was the transition from pediatric to adult care for CCS in NL. The boundaries of this case include perspectives from health care providers involved in the care of CCS. This includes pediatric providers, adult providers, and allied health providers. We sought to specifically examine the NL geographic region. Finally, data collection was completed using semi-structured interviews and collection of any additional information provided to CCS during the transition phase.

Inclusion/Exclusion Criteria

We used a purposive sampling technique for the recruitment of participants for this study (3). In consultation with the research team, it was decided that we wanted to reach as close to a comprehensive sample of participants involved in the care of CCS. As such, we sought to recruit pediatric oncologists, adult oncologist, family physician, nurse coordinator, and all allied health supports were selected for recruitment. With respect to inclusion of CCS, one member of the research team worked with the Pediatric Oncology Program to develop a list of 20 potential CCS who received care at the Janeway. This sample was chosen as a representative sample of CCS seen in the program. The CCS were from both rural and urban areas, experienced a variety of diagnosis, were diagnosed at different ages, and had varied levels of autonomy over their health care.

Ethical Considerations and Informed Consent

Ethics approval for the project was granted by the Newfoundland and Labrador Health Research Ethics Authority (4). When obtaining informed consent for the interviews with HCP, the interviewer (1) described the purpose of the study, (2) emphasized that the study was voluntary and that participants could withdraw at any point, (3) discussed any potential risk to taking part in the interview process, and (4) ensured participants of their anonymity. For all interviews conducted with CCS, an informed consent form was emailed to participants to be signed and returned prior to the interview. At the start of each interview, the interviewer read the following statements/questions to ensure that participants understood the consent process:

- Have you read the consent form?
- Do you have any questions or is there anything you would like to discuss about this study?
- Do you have enough information about the study?
- Do you understand that you are free to withdraw from the study at any time without giving a reason?
- It is your choice to be in the study and that you may not benefit directly from your involvement.

- Your privacy is protected and my records will be kept confidential.
- Do you agree to be audio taped?
- Do you agree to take part in this study?

Potential ethical considerations included possible PTSD related to childhood cancer diagnosis, treatment, and process. The research team discussed the potential need for supports if required.

Data Collection

Semi-structured interviews were the primary data collection tool used. However, we did ask participants to provide any information (i.e., survivorship care plan, educational material, etc.) that a CCS may receive during the transition process. Interview guides were developed and approved by the entire research team before their use. The goal of the interviews was to afford researchers the opportunity to investigate processes and barriers that were unique to the experiences of the CCS and HCP. As such, the questions focused on gaps that the research team previously identified. The interview guides were developed in advance of the interviews and were extensively reviewed by the research team to ensure appropriate language/terminology and assess the clarity of questions. The research team also focused on ensuring that the semi-structured approach was adhered to, in order to allow for CCS and HCP to explore issues in-depth. In an effort to build rapport and establish comfortable exchanges with the CCS we interviewed, the invitation letter was sent from the Janeway Oncology program. We also provided a summary of the research program in the invitation and described what to expect during the interview. Recognizing the importance of data triangulation, we attempted to collect data from multiple sources, including CCS, pediatric HCP, adult HCP, and allied health providers. Using these varied perspectives around the same issue should help develop a holistic understanding of the phenomenon of transitions for CCS. We also hoped to receive documentation provided to CCS during transition; however, we found that no information was provided to participants during their transition.

Data Analysis

We used qualitative descriptive and thematic analysis to analyze the interviews. This approach has been previously been used to study healthcare processes (5). Descriptive analysis was used to identify the processes by which CCS transition from pediatric to adult care. This approach was used to glean a factual summary of these processes.

Qualitative descriptive analysis was used to describe "transition from pediatric care to adult care." Qualitative description is a form of naturalistic inquiry with no specific assumptions about the data. The data is presented in the language of the participants. In this analysis, no attempt is made to present the data in a theoretical manner. The end result is a comprehensive summary of the transition process (5).

For the identification of barriers to improved transition, we used thematic analysis. We took the following steps in the thematic analysis process (5-7). We reviewed each interview transcript in full, for familiarization of the data, prior to beginning the coding process. Next, each transcript was manually coded by one researcher (DR). Review of the coding structure was completed by all members of the research team. Confirmation of the coding was completed by all members of the research team. After the coding was completed, we arranged the data by code. We organized the codes under “theme” headings (for example for the theme “Lack of Education Surrounding transitions,” several sub-codes are listed including understanding diagnosis, understanding treatment, understanding need for aftercare, education needs to start earlier, etc.). Once all main themes were identified, the researcher team reviewed the transcripts again for confirmation of themes. The research team also reviewed the transcripts after coding was complete to ensure that themes accurately represented the data. Field notes were also taken; however, these were not factored into the analysis as they did not add any additional information.

Results

Each theme emerged from a series of sub-themes. For example, "Challenges for Rural Patients" included the following sub-themes: limitations with respect to allied health support; lack of speciality care; the need for a new way for aftercare; challenges of travelling clinics; and added burden of coordination of care. The theme "Changes in Availability of Services" included: differences in how services were provided in pediatric and adult care; challenges accessing allied health services in the adult system;

long-wait times; added cost of services; lack of preparation; and dealing with a new structure of care. The sub-themes that were heard under the theme "Challenges with Navigating the System" included: unclear contact times after leaving pediatric care; minimal tools for navigating the adult health care system; and challenges with patient autonomy (because of nature of illness and comfort from pediatrics). Lastly, the theme "Lack of Education" was heard across all interviewees and was echoed across several questions posed to participants. It was found that interviewees discussed issues around: understanding diagnosis; treatments; aftercare; and screening/health promotion practices. It was clear that education around transition is needed earlier in pediatric care and that healthcare providers did not receive education specific to transition. Finally, a lack of formal preparation was felt from HCP and CCS. No documentation or survivorship care plan was provided to CCS. While each of these themes were strongly identified in the coding process, "Lack of Education" was more pervasive across different questions.

Chapter 5: Evaluating a Transition Workbook for Childhood Cancer Survivors: A Pilot Study

The Intervention

This workbook includes three sections. The first section is informational. The introduction is followed by a section on why transition into adult care matters. This section introduces the idea of transition, aftercare, and late effects. Next, is a description of the need for a new provider in adult care and some goals for transition are discussed. We also highlight the expectations a CCS should have for their adult aftercare team.

Section B is focused on management transition into adult aftercare and ensuring that CCS and their families are adequately educated on their current and future aftercare needs. This section was designed to be interactive. The Medical Record was meant to help CCS and their families learn more about their diagnosis, treatment, and future care. "My Healthcare Team" was created to ensure CCS had proper contact information for their healthcare providers in both the pediatric and adult aftercare settings. Following this section, was a topics checklist. This section lists important terms for CCS and leaves some blank spaces for additional terms specific to individual survivors. The Transition Readiness Assessment was developed by Got Transition and is designed to be completed with the assistance of healthcare providers to determine any area that could be improved during transition. The subsequent piece of this section was designed for CCS to be able to understand and describe their health in three sentences. The final interactive element to this section was the health passport. This is designed to house all pertinent information regarding a CCS previous cancer.

Section C of the workbook was geared towards providing additional information. This includes a section on health promotion tips and practical services and organizations. Based on the feedback of our study we intend to add a section specific to mental health in this section. We also intend to provide more local resources.

Overall, the specific actions that this workbook hopes to inspire include: patient autonomy, improved knowledge of cancer and treatment, improved knowledge of late effects, improved understanding of health promotion activities, and an understanding around navigating the adult healthcare system.

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Appendix B

Categorization of Models of Care for Childhood Cancer Survivors

Article	1. Which provider is primarily responsible for aftercare?	2. Which providers are regularly involved in providing aftercare?	3. Where is care provided?	4. How are survivors engaged?	5. Which services are provided?	6. Who receives services?
Aziz et al., 2011	• *Pediatrician*	• *Pediatrician or physician with "late effects" experience* • *Social worker* • *Psychologist* • *Nurse* • *Administrative staff*	• *Children's hospital* • *Children's cancer centre*	• *N/A*	Core health services were fairly uniform including surveillance for disease reoccurrence, screening for late effects, education, counselling, and social services	• *Survivors of all childhood cancer types.* • *64% provided CCS of all ages. Some provided care to CCS younger than 25 years.* • *2 years after the completion of cancer therapy or 5 years after their cancer diagnosis*
Kenney et al., 2011	• *Pediatrician*	• *Pediatric oncologist, nurse practitioner* • *PCP* • *Registered nurse* • *Mental health provider* • *Pediatric sub-specialist*	• *Pediatric oncology clinics*	• *Using electronic databases to capture survivor's data*	Educational activities, treatment summary, follow-up care plan, and survivor research	• *Primarily CCS 2-3 years off of treatment or 5 years after completion of treatment.* • *Some programs do not have a specific age after which CCS could not be seen in the*

Article	1. Which provider is primarily responsible for aftercare?	2. Which providers are regularly involved in providing aftercare?	3. Where is care provided?	4. How are survivors engaged?	5. Which services are provided?	6. Who receives services?
						pediatric program - *Other programs have age maximums ranging from 21 to 40 years.*
Michel et al., 2009	- *Pediatrician*	- *CCS who were surveyed preferred consultant-led* - *Pediatric oncologist and late effects nurse follow-up although they were open to other models of care*	- *Late effects clinic in Sheffield, UK.*	- *Risk stratification depending on type of treatment.* - *Could be postal or telephone up to more invasive follow-up*	- *Psychological support* - *Support groups* - *Nutritional information* - *Employment/career support.*	- *Diagnosis of any childhood cancer (less than 16 years) more than 5 years since diagnosis and currently registered in late effects clinic*
Oeffinger et al., 1998	- *Pediatrician*	- *Pediatric oncologist* - *Adult oncologist* - *Nurse practitioner/clinicians*	- *Over half of COG centres surveyed have a LTFU clinic at their institution*	- *Formal database exists in about 15% of COG institutions surveyed*	- *Educational materials* - *Preventative healthcare components*	- *COG institutions that have a mechanism for following up CCS over the age 18 years*
Oeffinger et al., 2004	- *Pediatrician*	- *N/A*	- *19.2% at cancer centre*	- *CCSS using last available address*	- *General physical examination*	- *CCS who have survived for 5 or more years.* - *One of the eight primary cancer groups and less*

Article	**1. Which provider is primarily responsible for aftercare?**	**2. Which providers are regularly involved in providing aftercare?**	**3. Where is care provided?**	**4. How are survivors engaged?**	**5. Which services are provided?**	**6. Who receives services?**
						than 21 years of age at diagnosis
Sedak et al., 2015	• *Pediatrician*	• *Pediatric oncologist* • *Pediatric oncology* • *Nurse practitioner* • *Internal medicine physician* • *Social worker* • *Neuropsychologist* • *Program manager* • *Database manager* • *Administrative support*	• *Childhood cancer centre at University Medical centre*	• *Cancer survivorship database maintained by the pediatric haematology/oncology department*	• *N/A*	• *Survivors of childhood cancer with 5 or more from their cancer diagnosis*
Earle et al., 2005	• *PCP* • *Nurse*	• *PCP* • *Nurse*	• *N/A*	• *N/A*	• *Would like to see more psychological support, insurance support, educational material*	• *CCS who were off treatment for at least 5 years*
Nathan et al., 2008	• *Pediatrician, distant follow-up* • *Nurse* • *PCP*	• *N/A*	• *Most received care in cancer centre*	• *CCSS by using healthcare records from 26 institutions*	• *N/A*	• *CCS with cancer before the age of 21 who were alive at least 5 years after their original diagnosis*
Nathan et al., 2013	• *PCP*	• *PCP; however, majority would prefer to work with others including in consultation with cancer centre*	• *Primary care setting*	• *N/A*	• *Survivorship care plan*	N/A

Article	1. Which provider is primarily responsible for aftercare?	2. Which providers are regularly involved in providing aftercare?	3. Where is care provided?	4. How are survivors engaged?	5. Which services are provided?	6. Who receives services?
Ristovski-Slijepcevic et al., 2009	• *Pediatrician*	• *Adult oncologist* • *PCP* • *Pediatric oncologist* • *Nurse* • *Social worker* • *Clinical psychologist Neuropsychologist* • *Dietician*	• *Pediatric healthcare settings across Canada*	• *Engaged through healthcare institution*	• *Survivorship care plan* • *Transitional planning*	• *Across Canada, the guidelines vary.* • *Although, many pediatric programs will keep survivors because of the absence of resources for adults.* • *Aftercare generally starts two or more years after the completion of active treatment*
Mertens et al., 2004	• *Pediatrician* • *PCP*	• *Multi-disciplinary teams using hybrid models of care*	• *Primary care* • *Tertiary care setting*	• *N/A*	• *Survivor education* • *Health insurance/access* • *Initiatives are important*	• *N/A*
Parkes et al., 2008	• *Phone/Text/E-mail*	• *None*	• *Follow-up through postal mail*	• *Engaged CCS who do not take part in aftercare*	• *None*	• *CCS who have survived cancer for 5 years of more and who have not attended clinical follow-up*
van Laar et al., 2013	• *Adult oncologist*	• *N/A*	• *CCS from pediatric and adult*	• *Engaged through Yorkshire cancer network and Humber*	• *N/A*	• *Survivors over the age of 18 that were*

Article	1. Which provider is primarily responsible for aftercare?	2. Which providers are regularly involved in providing aftercare?	3. Where is care provided?	4. How are survivors engaged?	5. Which services are provided?	6. Who receives services?
	- *Pediatric oncologist*		*oncology centres*	*and Yorkshire Coast Cancer Network*		*diagnosed before their 18th birthday and were at least 5 years post completion of treatment.*
Meacham et al., 2012	- *PCP (shared-care)*	- *PCP* - *Non-oncology providers*	- *N/A*	- *N/A*	- *Survivor healthcare plan*	- *N/A*
Blaauwbroek et al., 2008	- *Pediatrician* - *PCP*	- *Family physician* - *Pediatric oncologist*	- *University Medical Centre Groningen, Netherlands*	- *CCS chosen by use of computer program and recalled to LTFU*	- *Booklet* - *Summary of diagnosis* - *Treatment received* - *Potential late effects*	- *CCS 18 years and older who had been treated at the pediatric oncology department at least 5 years previously and were not involved in any childhood follow-up programs.*
Blaauwbroek et al., 2007	- *Pediatrician* - *PCP*	- *Family physician* - *Pediatric oncologist*	- *Primary care*	- *N/A*	- *N/A*	- *N/A*
Vetsch et al., 2016	- *Pediatrician* - *PCP*	- *Pediatrician* - *PCP*	- *Switzerland CCS population* - *CCS receiving care at university and regional hospital*	- *Engaged through the Swiss Childhood Cancer Registry.*	- *N/A*	- *CCS aged 11-17 years (survey of parents about their follow-up)*

Article	1. Which provider is primarily responsible for aftercare?	2. Which providers are regularly involved in providing aftercare?	3. Where is care provided?	4. How are survivors engaged?	5. Which services are provided?	6. Who receives services?
Signorelli et al., 2017	• *Pediatrician* • *Oncologist* • *Nurse*	• *Oncologist* • *Psychologist* • *Fertility specialist* • *Multidisciplinary team*	• *LTFU clinics that have been established in hospitals in ANZ.* • *Five hospitals ran outreach clinics in rural/remote locations.*	• *N/A*	• *COG guidance* • *SIGN*	• *Varies by clinic.* • *Range from <16 years to no limit.* • *Very flexible.*
Szalda et al., 2016	• *Adult Oncologist* • *PCP* • *Hybrid care*	• *Shared care* • *Both specialized survivorship clinic and a PCP and subspecialists.*	• *CCS transferred from Cancer Survivorship Program at the Children's Hospital of Philadelphia to adult-focused follow-up care in the past 1-5 years.*	• *Authors acknowledge that engagement is lacking and options for engagement should be explored.*	• *Completed measures of engagement with cancer-related follow-up* • *Other healthcare utilization*	• *CCS who have been transferred from pediatric survivorship care in prior 1-5 years.*
Lie et al., 2017	• *PCP* • *Oncologists,* • *Hybrid care*	• *PCP* • *Oncologist,* • *Psychologist* • *Specialists*	• *National university hospital in Norway*	• *Engaged though previous research two years prior using Norwegian Cancer registry*	• *Psychologist* • *Late-effects related screening*	• *CCS of Hodgkin and non-Hodgkin lymphoma in Norway*
Michel et al., 2017	• *Medical oncologist* • *Pediatricians*	• *Medical oncologist* • *Pediatricians* • *General practitioner* • *Pediatric oncologist.*	• *Switzerland hospital or own group practice*	• *Engaged through the Swiss Childhood Cancer Registry.*	• *N/A*	• *CCS in Switzerland*

Article	**1. Which provider is primarily responsible for aftercare?**	**2. Which providers are regularly involved in providing aftercare?**	**3. Where is care provided?**	**4. How are survivors engaged?**	**5. Which services are provided?**	**6. Who receives services?**
	• *General practitioner* • *Pediatric oncologist.*					
Casillas et al., 2017	• *Other*	• *N/A*	• *N/A*	• *Recruited through UCLA pediatric Hematology/Oncology Survivorship Database*	• *N/A*	• *CCS from greater Los Angeles area between 15-39 years of age with cell phone with text messaging capacity*

Appendix C

Interview Guide for CCS and HCP

Pediatric Oncologist Interview Guide

1. Could you describe your role as a healthcare provider?
2. Could you describe the process by which survivors of childhood cancer usually transition from your program into adult care?
    a. At what age do patients usually transfer into adult care?
    b. To whom do you usually transfer patients?
3. How do you currently prepare your patients for the transition into adult care?
    a. At what age do you start to prepare your patients?
4. What concerns/challenges do patients raise to you about their transition to adult care before they are transferred?
5. How do you think your current process for transferring patients from your clinic is working?
6. Are there added difficulties in transition for survivors in rural areas?
7. Our pediatric oncology program has a high retention rate for survivors. Why do you think the program been able to maintain such high retention rates?
8. What do you believe the goals of transition should be?
9. In your experience, do your patients experience changes in their availability to services due to their transition into adult care?
10. What suggestions do you have for improving the transferring of your patients into adult care?

11. What model of care/aftercare do patients currently experience?
    a. What do you think would work best here in Newfoundland?
    b. If no structure:
        i. Does there need to be a structured program in Newfoundland?
12. Given the competing ways to improve patient care, how would you rank improving the transition to adult care as a priority for your program?
13. Would you be interested in further training in this area? And if so, in what format?
14. Do you provide patients the tools/information to navigate the healthcare system as they age?
15. Is there anything else that you would like to add?
16. Do you have any documents related to transition that you could share?

Nurses/Allied Health Professionals Interview Guide

1. Could you describe your role as a healthcare provider?
2. Could you describe the process by which survivors of childhood cancer usually transition from your program into adult care?
    a. At what age do patients usually transfer into adult care?
    b. To whom do you usually transfer patients?
3. How do you currently prepare your patients for the transition into adult care?
    a. At what age do you start to prepare your patients?
    b. Does your organization give guidance of how the transition process should occur?
4. What concerns/challenges do patients raise to you about their transition to adult care before they are transferred?
5. How do you think your current process for transitioning patients from your clinic is working?
6. Are there added difficulties in transition for survivors in rural areas?
7. The Janeway pediatric oncology program has a high retention rate for survivors. Why do you think the program been able to maintain such high retention rates?
8. What do you believe the goals of transition should be?
9. Are you aware of any concerns regarding patients transition to adult care?
    a. Are you aware of any challenge's patients experience after transitioning?
    b. What do you see as the enablers and barriers of a good transition?

    c. In your experience, do your patients experience changes in their availability to services due to their transition into adult care?

10. What suggestions do you have for improving the transferring of your patients into adult care?
11. What model of care/aftercare do patients currently experience?
    a. What do you think would work best here in Newfoundland?
    b. If no structure:
        i. Does there need to be a structured program in Newfoundland?
12. Given the competing ways to improve patient care, how would you rank improving the transition to adult care as a priority for your program?
13. Do you feel you were adequately trained to best assist your patients as they reach the point on transition?
    a. Would you be interested in further training in this area? And if so, in what format?
14. Do you provide patients the tools/information to navigate the healthcare system as they age?
15. Are there any resources or supports that you would like to see developed to better support either patients or providers?
16. Is there anything else that you would like to add?
17. Do you have any documents related to transition that you could share?

Family Physicians Interview Guide

1. Could you describe your role as a healthcare provider?
    a. What is your role as it relates to childhood cancer survivors?
    b. Experience with childhood cancer survivors?
2. Could you describe the process by which survivors of childhood cancer usually transition into your program?
    a. At what age do patients usually transfer into adult care?
3. Approximately how many survivors of childhood cancer do you currently have in your practice?
    a. How many would you typically have transfer to your practice in a year?
4. At what age do patients usually transfer into your practice for the care of their cancer?
5. Does your organization give physicians guidance of how the transition process should occur?
6. The Janeway Oncology program has had high retention rates in their aftercare program. Why do you believe the pediatric oncology program has been able to maintain such high retention rates?
7. What do you believe the goals of transition should be?
8. What do you see as enablers and barriers of good transition?
9. Do you feel survivors of childhood cancer are usually prepared to make the transition when they do?

10. How well do you think the current model of transition working?
11. How do you think the current process for transferring patients from your clinic is working?
12. What concerns do patients raise to you about their transition to adult care?
13. In your experience, what challenges do patients experience after transitioning?
14. In your experience, do your patients experience changes in their availability to services due to their transition into adult care?
15. What model of care/aftercare do patients currently experience? (Provide background if needed)
    a. What do you think would work best here in Newfoundland?
    b. If no structure:
        - Does there need to be a structured program in Newfoundland?
16. What suggestions do you have for improving the transitioning of survivors of childhood cancer into adult care in our province?
17. Do you feel you were adequately trained to best assist your patients as they reach the point on transition?
18. Would you be interested in further training in this area? And if so, in what format?
19. Do you provide patients the tool to navigate the healthcare system as they age?
20. In your experience, do your patients experience difficulties with navigating the healthcare system
21. Is there anything else that you would like to add?
22. Do you have any documents related to transition you could share?

Childhood Cancer Survivors Interview Guide

1. Could you provide me with some details of your;
    a. Cancer diagnosis
    b. Treatment
2. When did you complete your treatment?
3. How old were you when you;
    a. Finished pediatric care?
    b. Transferred into adult care?
4. Could you describe what happened when you finished your care at the Janeway
    a. Pediatric aftercare?
    b. How long was care provided at the Janeway?
5. After your time at the Janeway, what did your care for your past cancer diagnosis look like?
6. Are you aware of any aftercare?
    a. Are you aware of any guidelines for survivors?
7. Was there any conservation about a transition from the Janeway into adult care?
8. What do you believe the goals of transition should be?
9. How well is the current transition model meeting these goals?
10. What was the process by which you were transferred to an adult physician?
11. What did your pediatric care team do to prepare you for the transition into adult care?

12. Did you feel prepared to make the transition when you did?
13. Who is responsible for your long-term follow-up?
    a. Who were you referred to for your adult aftercare?
14. Who are you currently seeing for any cancer related issues?
15. How do you feel about how you were transferred into adult care?
16. Do you have any concerns about your transition to adult care?
17. Did you experience changes in availability of services due to your transition into adult care?
    a. If so, how are you dealing with the loss of services?
18. What type of aftercare do you think would work best here at the HSC?
19. Where are you in your aftercare (explain, if needed)?
20. What is your understanding of the point of aftercare?
21. What do you see as enablers for a good transition?
    a. Barriers of good transition?
22. What suggestions do you have for improving the transition into adult care for survivors of childhood cancer?
23. How do you find navigating the healthcare system?
    a. Do you have any challenges?
    b. Has anyone helped you?
24. Is there anything else that you would like to add?

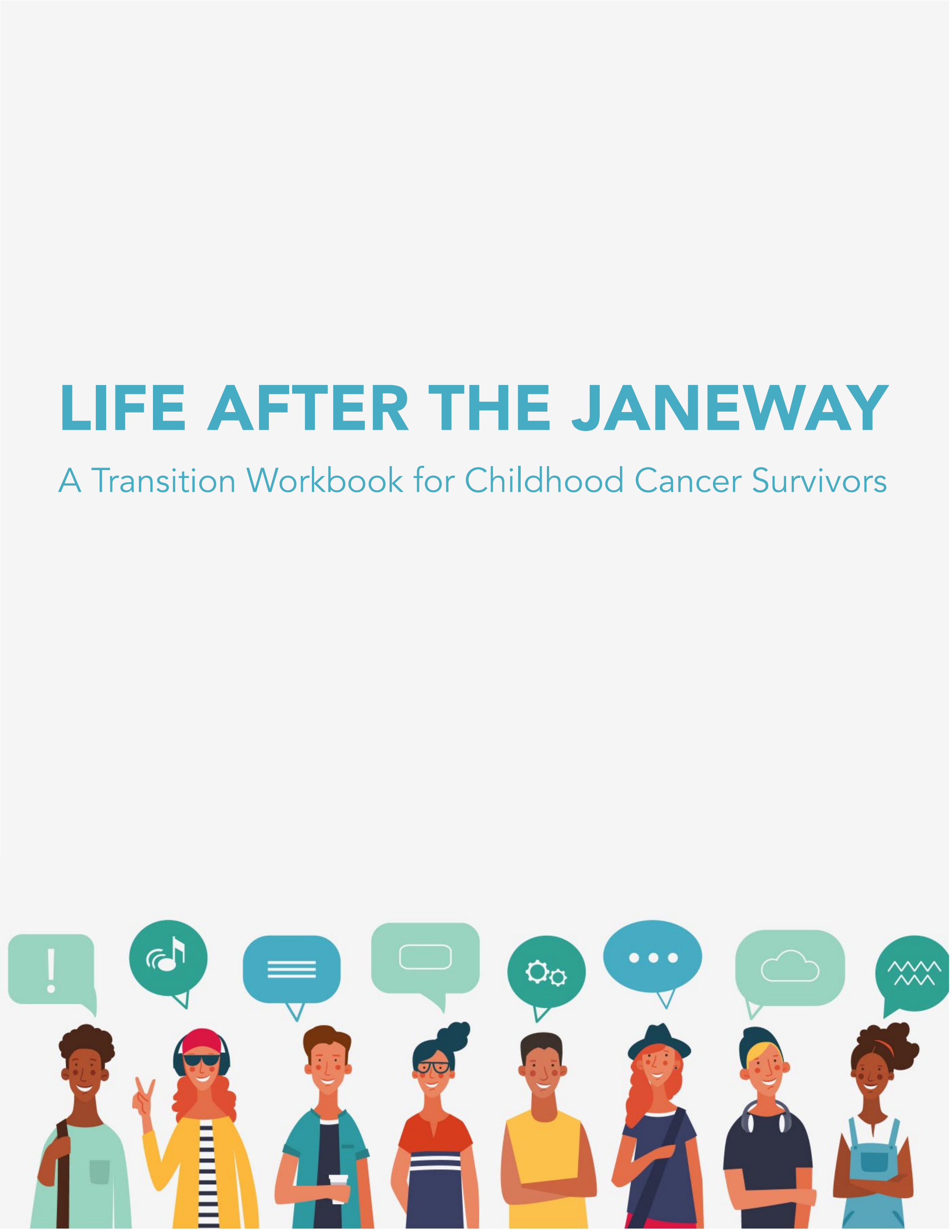
Adult Provider Interview Guide

1. Could you describe your role as a healthcare provider?
2. Could you describe the process by which survivors of childhood cancer usually transition from your program into adult care?
    - At what age do patients usually transfer into adult care?
    - To whom do you usually transfer patients?
3. Approximately how many survivors of childhood cancer transition into your practice?
4. At what age do patients usually transfer from your practice into adult care?
5. Does your organization give guidance of how the transition process should occur?
6. Why do you believe the pediatric oncology program has been able to maintain such high retention rates?
7. What do you believe the goals of transition should be?
8. How well is the current transition model meeting your goals and your patients' goals?
9. How do you feel survivors of childhood cancer are usually prepared to make the transition when they do?
10. What do you see as the enablers and barriers of a good transition?
11. How do you think your current process for transitioning patients into your clinic is working?
12. Are you aware of any challenge's patients experience after transitioning?

13. In your experience, do your patients experience changes in their availability to services due to their transition into adult care?
14. What type of MOC/aftercare do you think would work best here at the HSC? (Provide explanation if needed)
15. What suggestions do you have for improving the transferring of your patients into adult care?
16. Do you complete any professional development and/or education on caring for adult survivors of childhood cancer?
17. What supports and/or resources are in place for adult providers to care for survivors of childhood cancer?
18. Do you feel you were adequately trained to help with the transition process?
19. In your experience, do your patients experience difficulties with navigating the healthcare system?
20. Is there anything else that you would like to add?
21. Do you have any documents related to transition you could share?

Appendix D

"Life After the Janeway" Workbook



LIFE AFTER THE JANEWAY

A Transition Workbook for Childhood Cancer Survivors

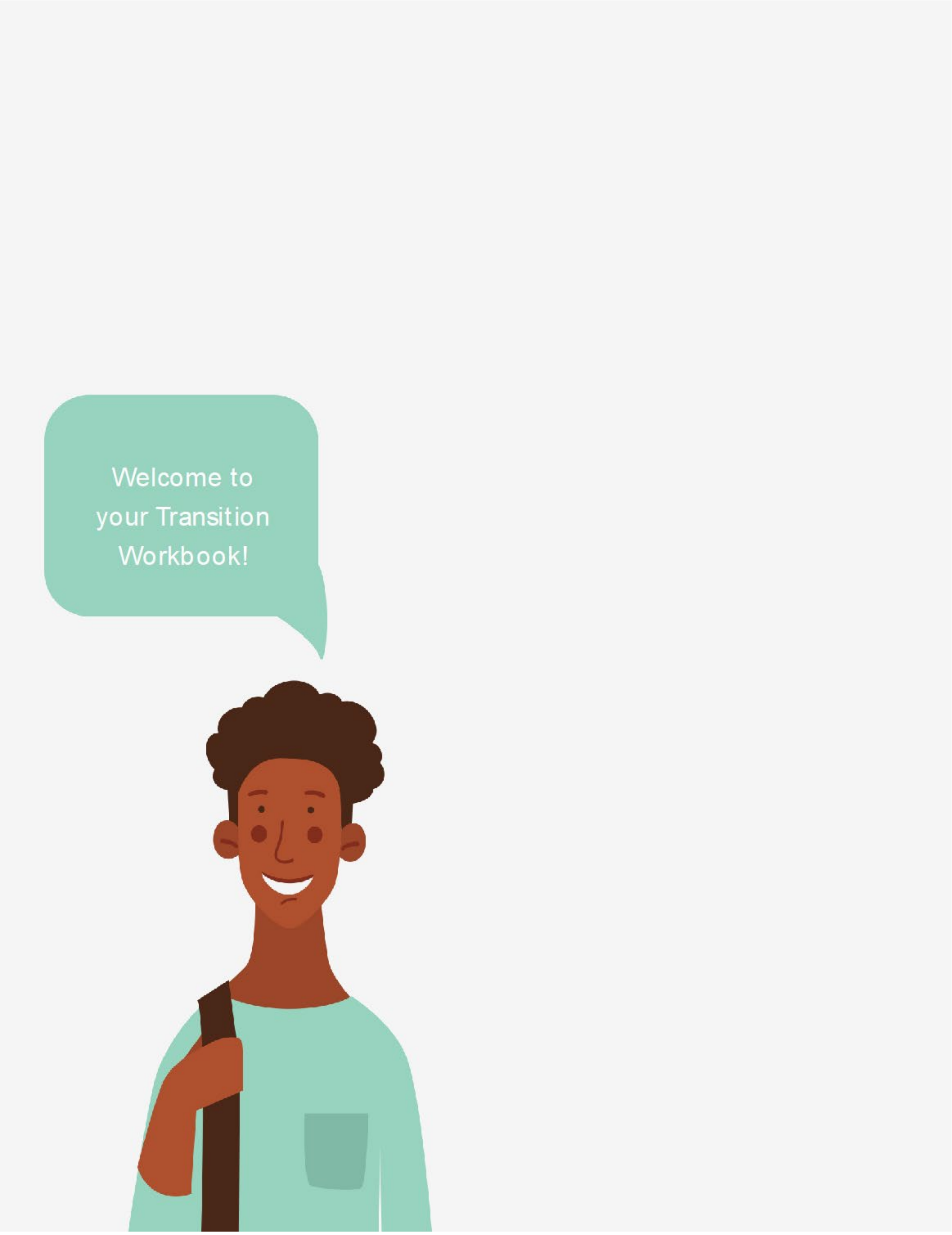

Welcome to your Transition Workbook!

TABLE OF CONTENTS

WELCOME

WELCOME TO YOUR PERSONALIZED TRANSITION WORKBOOK

As a person who survived cancer in childhood, you have been followed after your treatment by the Janeway Oncology program. This post-treatment follow-up care, which is called aftercare, is crucial for watching for any effects that your cancer or its treatment might have on your health, and for making sure that you receive the right treatments for those effects. As you become a young adult, this follow-up care is still important.

This workbook provides you and your family with practical information to help you transition from the Janeway Oncology program into adult care. This workbook has been developed by researchers at Memorial University with the help of childhood cancer survivors, pediatric health care providers, and adult health care providers.

The workbook begins by providing information about cancer treatments and the importance of aftercare. The second section is designed to be an interactive workbook for you to complete and review with your health care providers. The final section provides information about resources that you may find helpful. We encourage you to complete this workbook and customize it with your own information. Please keep this workbook in a secure location. If you choose to fill out your health details you will be responsible for the privacy and confidentiality of the information.

We appreciate that transitioning from the Janeway to another provider can be a difficult time. We hope that this workbook will be a helpful support to you during this process.

Kind regards,
Research team

SECTION A: INFORMATION

INTRODUCTION

This section is designed to help you understand the value of **aftercare** and the importance of successful transition to adult care. It contains information on important things to keep in mind, things to watch out for, and things to expect as you transition to adult-focused health care.

Aftercare is important for a number of reasons:

- It provides on-going health care
- It can identify effects that your cancer treatment may have on your health
- It gives you up to date information about your health needs
- It can provide referrals to other services when needed
- It can improve your quality of life and well-being

WHY DOES MY TRANSITION TO ADULTCARE MATTER?

Over the last 30 years, there have been huge improvements for children diagnosed with cancer. Survival rates for childhood cancer have reached over 80%. These improvements are due to improved cancer therapies. These therapies can have effects on health, these are called late effects. In fact, upwards of two-thirds of survivors of childhood cancer will face some form of late effect from their cancer treatment[1, 2]. Adult survivors of childhood cancers are at higher risk of developing second cancers, problems with the heart, kidneys, lungs, and other organs, increased body weight, educational difficulties, mental health issues such as medical post-traumatic stress, and reduced fertility[1, 2]. A specific person's risks depend on what treatment they had. It is important that a survivor knows the risks of their treatment.

Far too often survivors finish their care at their pediatric institutions (i.e. the Janeway) and do not take part in continued cancer-specific adult aftercare. Making sure the transition from pediatric care to adult care is supported can help with continued care into adulthood. Outside of healthcare, the transition from adolescence to adulthood is a time where you will experience a lot of growth and change. This complex time in your life is further complicated by making the transition from the supported system of pediatric care into the adult system where you will be expected to take more responsibility for organizing your own care. Transitioning into adult care is important for reducing the risk of developing long-term complications and is strongly encouraged in long-term follow-up guidelines for childhood cancer survivors[3-5].

WHY DO I NEED TO TRANSITION TO A NEW PROVIDER?

As a young adult, your health care, including your aftercare, should be delivered by a provider with expertise in adults. This can be exciting time in your life, when you take on more responsibility for your healthcare. But it can also be challenging. We hope that this workbook can help guide you through this journey.

WHAT DOES A SUCCESSFUL TRANSITION LOOK LIKE?

Transition will be different for every person[3-5], but some basic goals for transition include:

- Transition planning that is individualized
- Transition planning that starts before the transition
- Patients developing independence with their healthcare
- No gaps in care between pediatric and adult care
- Coordination between the patient, pediatric health care team, and adult health care team
- Patients learn how to communicate with their healthcare providers
- Patients understand their past diagnosis, treatment history, and risk of late effects

WHAT SHOULD I EXPECT FROM MY ADULT AFTERCARE TEAM?

There will be some changes in your care from the pediatric to adult care setting. The following are some new expectations:

1. It is now your responsibility to make sure you attend appointments and receive the care you need. If you have not already taken control over your care, now is the time to start!
2. You might not be reminded of appointments and follow-ups. It may be up to you to ensure your continued care is a priority.
3. Your healthcare team will likely speak to you, instead of your parents, during visits.
4. The focus of your aftercare will be health promotion and monitoring for potential late effects.
5. Your follow-up will be focused on making sure you don’t have any early signs of late effects based on your past treatment.
6. Your family doctor can continue to be your primary caregiver and can provide support through your aftercare.
7. Your aftercare provider will help guide you on the specifics of you care aftercare journey.

SECTION B: MANAGEMENT

This section is designed to help you prepare to transition out of the Janeway and into adult care. We hope that you can use this booklet to learn more about your diagnosis, treatment and future care. Follow the instructions in each segment of this booklet and fill out the required fields to help you keep track of your transition journey.

MY MEDICAL RECORD

Name: ____ Date of Birth (d/m/y): _______________

Age at diagnosis: ____ Therapy start date (d/m/y): __________ Therapy end date (d/m/y): ________

Major complications of therapy: ____
____
____

Chemotherapy drugs: ____
____
____

Site(s) of radiation: ____

Late effects to watch for: ____

MY HEALTH CARE TEAM

Fill out the following information on your healthcare team and keep it for your records.

My Pediatric Care Team

Oncologist/ Staff Physician: ____________________

Number: ____________ Email: ____________

Contact Nurse: ____________________

Number: ____________ Email: ____________

Social Worker: ____________________

Number: ____________ Email: ____________

Other Important Names and Numbers

Name/title (e.g.. My radiation oncologist, my psychologist):

Name/title: ____________________

Number: ____________ Email: ____________

Name/title: ____________________

Number: ____________ Email: ____________

Name/title: ____________________

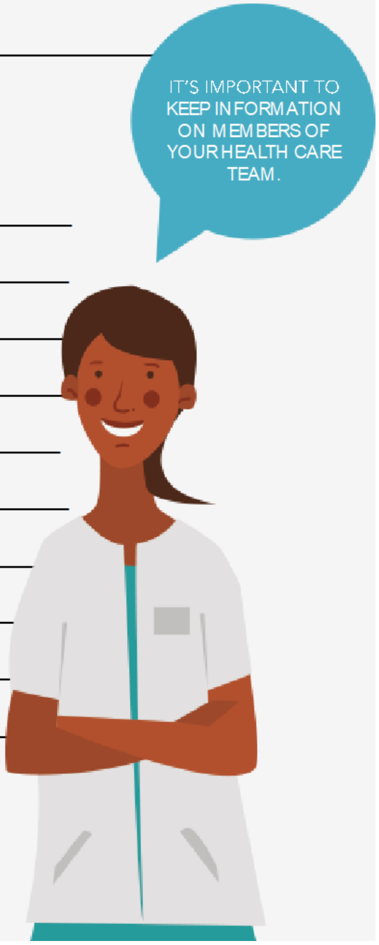
Number: ____________ Email: ____________

Name/title: ____________________

Number: ____________ Email: ____________

Name/title: ____________________

Number: ____________ Email: ____________



My Adult Care Team

*Oncologist/ Staff Physician:*________________________________

Number:__________________ Email: __________________________

Contact Nurse: ____

Number:__________________ Email: __________________________

Social Worker: ____

Number:__________________ Email: __________________________

Other Important Names and Numbers

Name/title (e.g.. My radiation oncologist, my psychologist):

Name/title: ____

Number:__________________ Email: __________________________

Name/title: ____

Number:__________________ Email: __________________________

Name/title: ____

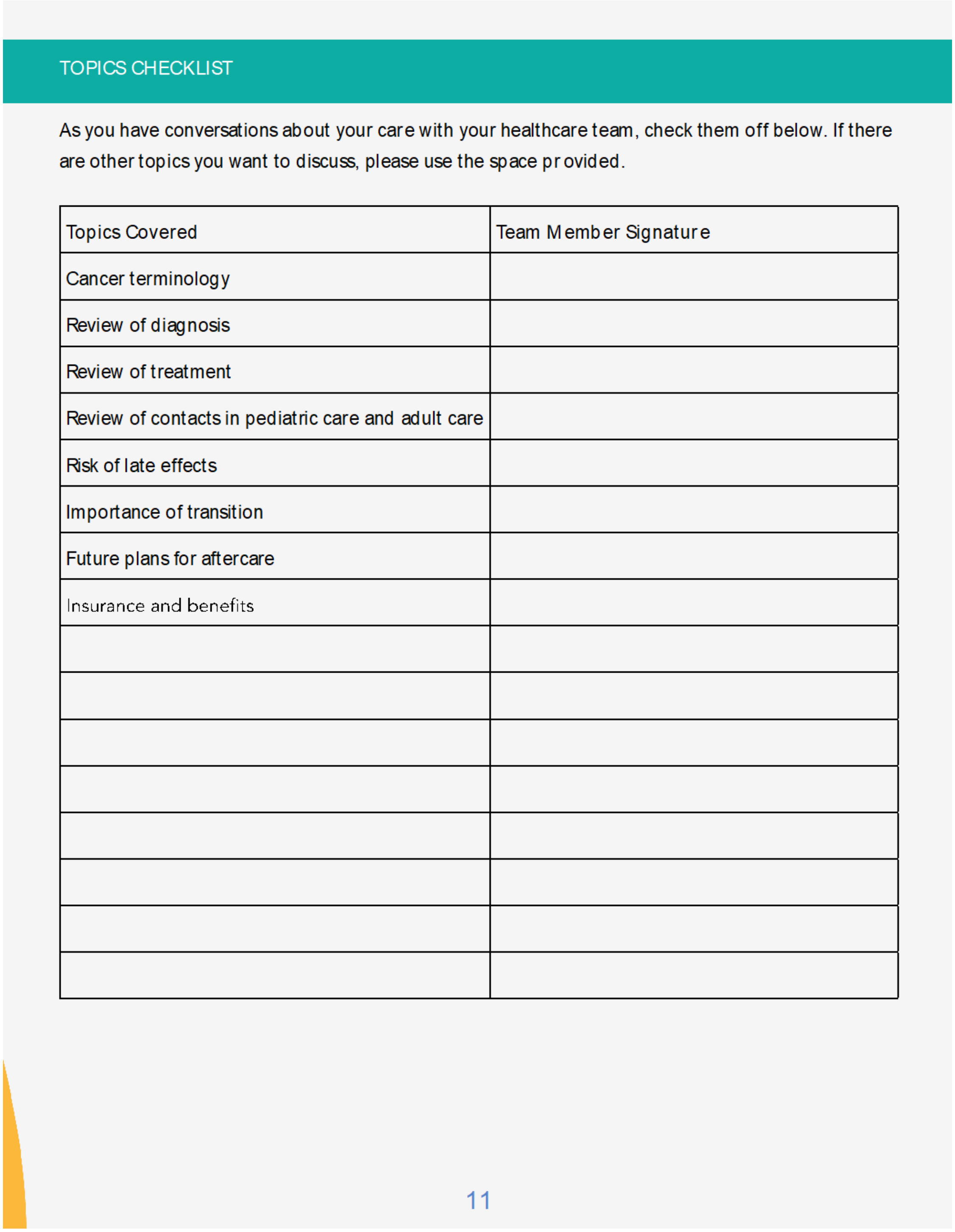
Number:__________________ Email: __________________________

Name/title: ____

Number:__________________ Email: __________________________

Name/title: ____

Number:__________________ Email: __________________________



TOPICS CHECKLIST

As you have conversations about your care with your healthcare team, check them off below. If there are other topics you want to discuss, please use the space provided.

Topics Covered	Team Member Signature
Cancer terminology	
Review of diagnosis	
Review of treatment	
Review of contacts in pediatric care and adult care	
Risk of late effects	
Importance of transition	
Future plans for aftercare	
Insurance and benefits	

ARE YOU READY TO TRANSITION TO ADULT CARE?

This checklist has been developed using research with childhood cancer survivors, pediatric healthcare providers, and adult healthcare providers. We have adapted this transition readiness tool with the permission of "got transition." Fill out the checklist with the statement that best describes you. Once you are done, go over the table with your health care provider to identify anything you feel you still need to work on.

Transition Readiness Assessment

Transition Importance and Confidence

On a scale of 0 to 10, please circle the number that best describes how you feel right now.

How important is it to you to prepare for/change to an adult doctor?										
0 (not)	1	2	3	4	5	6	7	8	9	10 (very)
How confident do you feel about your ability to prepare for/change to an adult doctor?										
0 (not)	1	2	3	4	5	6	7	8	9	10 (very)

My Health

Please check the box that applies to you right now.

	Yes, I know this	I need to learn	Someone needs to do this… who?
I know my medical needs.			
I can explain my medical needs to others.			
I know my symptoms for which I need to seek medical care.			
I know what to do in case I have a medical emergency.			
I know my own medicines, what they are for, and when I need to take them.			
I know my allergies to medicines and medicines I should not take.			
I carry important health information with me every day (e.g. insurance card, allergies, medications, emergency contact information, medical summary).			

	Yes, I know this	I need to learn	Someone needs to do this… who?
I understand how health care privacy changes at age 18 when I legally become an adult.			
I can explain to others how my customs and beliefs affect my health care decisions and medical treatment.			
Using Health Care			
I know or I can find my doctor's phone number.			
I make my own doctor appointments.			
Before a visit, I think about questions to ask.			
I have a way to get to my doctor's office.			
I know to show up 15 minutes before the visit to check in.			
I know where to go to get medical care when the doctor's office is closed.			
I have a file at home for my medical information.			
I have a copy of my current plan of care.			
I know how to fill out medical forms.			
I know how to get referrals to other providers.			
I know where my pharmacy is and how to refill my medicines.			
I know where to get blood work or x-rays if my doctor orders them.			
I have a plan so I can keep my health insurance after 18 or older.			
My family and I have discussed my ability to make my own health care decisions at age 18.			

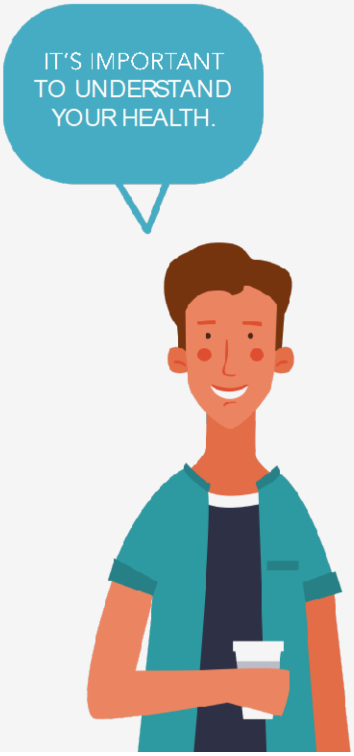
RECAP YOUR HEALTH HISTORY IN 3 STATEMENTS

Knowing your diagnosis, disease history and current health status are critical to manage your future health care needs. Being able to give your health summary will come in handy at future appointments. It shows that you understand your health.

STATEMENT 1: Your name, age and your main medical issues.
STATEMENT 2: Treatment, hospitalizations and your medications.
STATEMENT 3: Any current troubles/worries you are having.

Example:

"Hello, my name is Alex Anderson, I am 16 years of age and I am a childhood cancer survivor. I was diagnosed with leukemia at age 3. I received chemotherapy as treatment for my cancer. I was taking viscristine but I am no longer taking any medications. I do not have any current conditions but I am concerned about late effects of my cancer treatment as I get older."



Now, you give it try:

____
____
____
____
____
____
____
____
____
____
____

DON'T FORGET YOUR HEALTH PASSPORT WHEN YOU HEAD TO ADULTCARE

You're on your way to adult care! Don't forget to take your health passport with you! Work with your healthcare team to fill this out accurately. You can take your passport out of this workbook and keep it with you in your wallet. Your passport will give information about your pass cancer treatment such as past treatments, medication, and other critical information. You might also want to take a picture of your health passport with your smartphone in case it gets misplaced.

MY HEALTH PASSPORT	
Name	
DOB (dd/mm/yyyy)	
Cancer diagnosis	
Current medical problems	
Past treatment	
Current medications	
Operations	
Drug allergies	
Other allergies	
Specialist	
Primary care provider	
Insurance coverage	

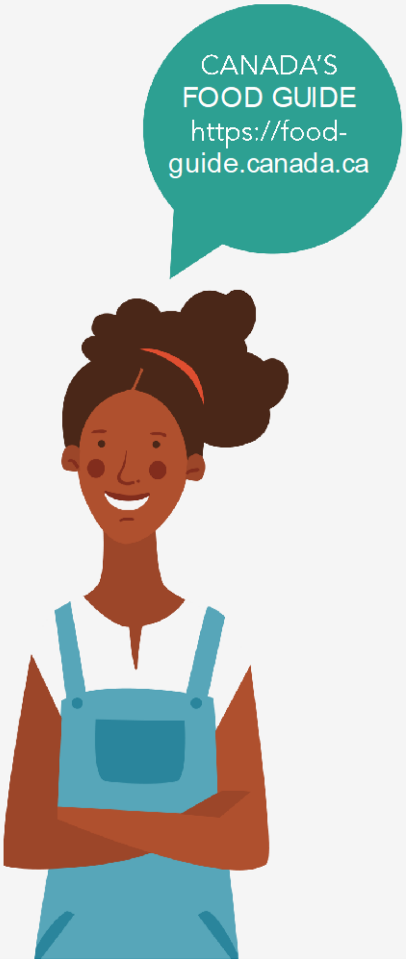
Adapted from https://www.sickkids.ca/myhealthpassport/FormPassport.aspx?FormId=3

SECTION C: RESOURCES AND ADDITIONAL INFORMATION

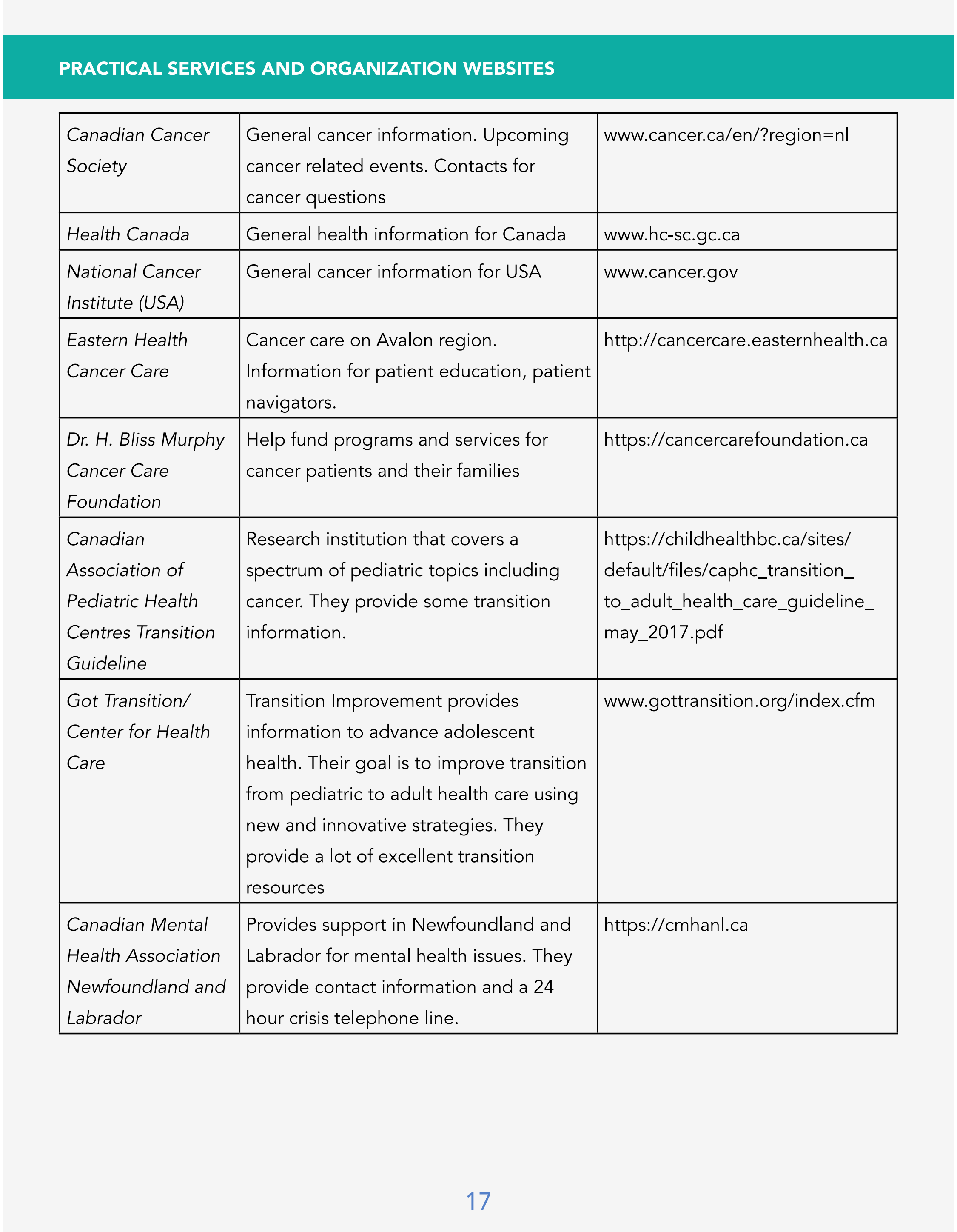
This section is designed to provide you with any additional information or resources you may need as you prepare for adult care. There is some space in this section for you to jot down any questions or notes you may have.

HEALTH PROMOTION TIPS

Continued health promotion is important regardless of your medical history. Below are some general health promotion practices important to everyday life [6,7].



- Care for your mental health
- Maintain a healthy body weight
- Embrace a physically active lifestyle
- Follow the Canada's Food Guide
- If you drink alcohol, limit consumption
- Attend follow-up appointments
- Take part in regular screening practices



PRACTICAL SERVICES AND ORGANIZATION WEBSITES

Canadian Cancer Society	General cancer information. Upcoming cancer related events. Contacts for cancer questions	www.cancer.ca/en/?region=nl
Health Canada	General health information for Canada	www.hc-sc.gc.ca
National Cancer Institute (USA)	General cancer information for USA	www.cancer.gov
Eastern Health Cancer Care	Cancer care on Avalon region. Information for patient education, patient navigators.	http://cancercare.easternhealth.ca
Dr. H. Bliss Murphy Cancer Care Foundation	Help fund programs and services for cancer patients and their families	https://cancercarefoundation.ca
Canadian Association of Pediatric Health Centres Transition Guideline	Research institution that covers a spectrum of pediatric topics including cancer. They provide some transition information.	https://childhealthbc.ca/sites/default/files/caphc_transition_to_adult_health_care_guideline_may_2017.pdf
Got Transition/ Center for Health Care	Transition Improvement provides information to advance adolescent health. Their goal is to improve transition from pediatric to adult health care using new and innovative strategies. They provide a lot of excellent transition resources	www.gottransition.org/index.cfm
Canadian Mental Health Association Newfoundland and Labrador	Provides support in Newfoundland and Labrador for mental health issues. They provide contact information and a 24 hour crisis telephone line.	https://cmhanl.ca

QUESTIONS TO ASK YOUR HEALTH PROVIDERS

1)____
2) ____
3) ____
4) ____
5) ____
6) ____
7) ____
8) ____
9____
10) ____

CHILDHOOD CANCER TRANSITION TERMINOLOGY

The main goal of transition from pediatric care to adult care is to receive continuous, coordinated care throughout the course of your life. You may hear the following terms as you journey from pediatric care to adult care:

Models of care: Program for cancer survivors that addresses health care needs in the context of the four categories of survivorship care components (prevention, surveillance, intervention, and coordination)

Aftercare: The care received after surviving cancer. Aftercare is dedicated to monitoring and managing possible late effects of pediatric cancer and can occur in a pediatric and/or adult setting.

Late effect: A late effect is a side effect of cancer treatment. Many people who have received treatment for cancer have a risk of developing late effects. Example of late effects include: heart problems, second cancers and emotional issues.

References

1. Friedman, D.L., D.R. Freyer, and G.A. Levitt, Models of care for survivors of childhood cancer.Pediatric blood & cancer, 2006. 46(2): p. 159-168.
2. Oeffinger, K.C., et al., Chronic health conditions in adult survivors of childhood cancer.New England Journal of Medicine, 2006. 355(15): p. 1572-1582.
3. Canadian Association of Paediatric Health Centres (CAPHC), A Guideline for Transition From Paediatric to Adult Health Care for Youth with Special Health Care Needs: A National Approach. CAPHC National Transitions Community of Practice, 2016.
4. Kaufman, M. and J. Pinzon, Transition to adult care for youth with special health care needs.Paediatr Child Health, 2007.12(9): p. 785-93.
5. American Academy of Pediatrics, American Academy of Family Physicians, & American College of Physicians-American Society of Internal Medicine, A consensus statement on health care transitions for young adults with special health care needs, 2002. Pediatrics, 110 (Supplement 3), 1304-1306.
6. Demark-Wahnefried, W. and L.W. Jones, Promoting a healthy lifestyle among cancer survivors.Hematology/oncology clinics of North America, 2008. 22(2): p. 319-342.
7. Canadian Cancer Society, Long-term Survivorship. 2019. Retrieved from https://www.cancer.ca/en/cancer-information/cancer-type/childhood-cancer-information/long-term-survivorship/?region=on

NOTES

Appendix E

Online Survey

Healthcare Provider Survey

Start of Block: Block 1

Evaluating "Life After the Janeway"- A Childhood Cancer Transition Workbook

"Life After the Janeway" is a new educational resource for Childhood Cancer Survivors preparing to move into adult care. In order to maximize its usefulness, we are asking for your assistance in completing this short evaluation survey. The survey evaluates the resource's clarity and usability. After reviewing the *"Life after the Janeway"* workbook, please complete the following survey, which should take between 5-10 minutes.   Thank you for your time!

End of Block: Block 1

Start of Block: Block 2

Which of the following do you identify as?

○ *Pediatric Healthcare Provider*

○ *Adult Healthcare Provider*

○ *Other, please specify below*

____

Which of the following do you identify as?

○ *Oncologist*

○ *Family Physician*

○ *Social Worker*

○ *Dietitian*

○ *Physiotherapist*

○ *Psychologist*

○ *Other, please specify below*

____

End of Block: Block 2

Start of Block: PEMAT-P

Section 1- Understandability

This section assesses whether the *"Life After the Janeway"* workbook is clear for individuals of diverse backgrounds and varying levels of health literacy. Please indicate if you agree or disagree with the following statements.

The purpose of the *"Life After the Janeway"* workbook is clear.

○ *Disagree*

○ *Agree*

The workbook does not include information or content that distracts from its purpose.

○ *Disagree*

○ *Agree*

The workbook uses common, everyday language. Medical terms are used only to familiarize audience with the terms. When used, medical terms are defined.

○ *Disagree*

○ *Agree*

Medical terms are used only to familiarize audience with the terms. When used, medical terms are defined.

○ *Disagree*

○ *Agree*

The workbook uses the active voice (e.g., *Allison changed the flat tire* versus *the flat tire was changed by Allison*)

○ *Disagree*

○ *Agree*

Numbers appearing in the workbook are clear and easy to understand.

○ *Disagree*

○ *Agree*

○ *N/A (No numbers)*

The workbook does not expect the user to perform calculations.

○ *Disagree*

○ *Agree*

The workbook breaks or "chunks" information into short sections.

○ *Disagree*

○ *Agree*

The workbook's sections have informative headers.

○ *Disagree*

○ *Agree*

The workbook presents information in a logical sequence.

○ *Disagree*

○ *Agree*

The workbook provides a summary.

○ *Disagree*

○ *Agree*

The workbook uses visual cues (e.g., arrows, boxes, bullets, bold, larger font, highlighting) to draw attention to key points.

○ *Disagree*

○ *Agree*

The workbook uses visual aids whenever they could make content more easily understood (e.g., illustration of healthy portion size).

○ *Disagree*

○ *Agree*

The workbook's visual aids reinforce rather than distract from the content.

○ *Disagree*

○ *Agree*

The workbook's visual aids have clear titles or captions.

○ *Disagree*

○ *Agree*

The workbook uses illustrations and photographs that are clear and uncluttered.

○ *Disagree*

○ *Agree*

○ *N/A (no visual aids)*

The workbook uses simple tables with short and clear row and column headings.

○ *Disagree*

○ *Agree*

○ *N/A (no tables)*

Page Break

Section 2- Actionability

This section assesses whether materials are actionable and individuals of diverse backgrounds and varying levels of health literacy can identify what they need to do based on the information presented.

The workbook clearly identifies at least one action the user can take.

○ *Disagree*

○ *Agree*

The workbook addresses the user directly when describing actions.

○ *Disagree*

○ *Agree*

The workbook breaks down any action into manageable, explicit steps.

○ *Disagree*

○ *Agree*

The workbook provides a tangible tool (e.g., menu planners, checklists) whenever it could help the user take action.

○ *Disagree*

○ *Agree*

The workbook provides simple instructions or examples of how to perform calculations.

○ *Disagree*

○ *Agree*

○ *N/A (calculations)*

The workbook explains how to use the charts, graphs, tables, or diagrams to take actions.

○ *Disagree*

○ *Agree*

○ *N/A (No charts, graphs, tables, or diagrams)*

The workbook uses visual aids whenever they could make it easier to act on the instructions

○ *Disagree*

○ *Agree*

End of Block: PEMAT-P

Start of Block: Block 3

How likely is it that you would use this workbook?

- ○ *Very Likely*
- ○ *Likely*
- ○ *Somewhat likely*
- ○ *Undecided*
- ○ *Somewhat Unlikely*
- ○ *Unlikely*
- ○ *Very Unlikely*

How likely would you be to recommend this workbook to an adolescent childhood cancer survivor who is about to transition into adult care?

- ○ *Very Likely*
- ○ *Likely*
- ○ *Somewhat likely*
- ○ *Undecided*
- ○ *Somewhat Unlikely*
- ○ *Unlikely*
- ○ *Very Unlikely*

Do you have any feedback for making this workbook more useful to survivors of childhood cancer or healthcare providers? If so, please use the space below to provide feedback.

Do have any concerns about this workbook? If so, please use the space below to describe them.

If you have any additional feedback on the workbook please provide it in the space below.

Thank you for your time!

Your contribution is much appreciated.

End of Block: Block 3

Childhood Cancer Survivors, Family Members and Community Group Survey

---

Start of Block: Block 1

Evaluating "Life After the Janeway"- A Childhood Cancer Transition Workbook

"Life After the Janeway" is a new educational resource for Childhood Cancer Survivors preparing to move into adult care. In order to maximize its usefulness, we are asking for your assistance in completing this short evaluation survey. The survey evaluates the resource's clarity and usability. After reviewing the *"Life after the Janeway"* workbook, please complete the following survey, which should take between 5-10 minutes to complete. Thank you for your time!

End of Block: Block 1

Start of Block: Block 2

Which of the following do you identify as?

○ *Childhood cancer survivor*

○ *Family member of a childhood cancer survivor*

○ *Member of community support group for childhood cancer survivors*

Skip To: Q69 If Which of the following do you identify as? != Childhood cancer survivor

When receiving treatment for your childhood cancer were you living in an urban or rural area?

○ *Urban*

○ *Rural*

Are you currently receiving aftercare for your childhood cancer?

○ *Yes*

○ *No*

Skip To: Q69 If Are you currently receiving aftercare for your childhood cancer? = No

Do you receive your aftercare in an urban or rural area?

○ *Urban*

○ *Rural*

What gender do you identify as?

○ *Male*

○ *Female*

○ *Other*

○ *Prefer not to answer*

What is the highest level of school you have completed or the highest degree you have received?

- ○ *Less than high school degree*
- ○ *High school graduate (high school diploma or equivalent including GED)*
- ○ *Some college but no degree*
- ○ *Bachelor's degree in college*
- ○ *Master's degree*
- ○ *Doctoral degree*
- ○ *Professional degree*

What is your age?

- ○ *18-24 years of age*
- ○ *25-34 years of age*
- ○ *35-44 years of age*
- ○ *45-54 years of age*
- ○ *55-64 years of age*
- ○ *65 years and older*

Which statement best describes your current employment status?

○ *Employed*

○ *Not currently Employed*

○ *Prefer not to answer*

End of Block: Block 2

Start of Block: PEMAT-P

Section 1- Understandability

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○ *Disagree*

○ *Agree*

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○ *Disagree*

○ *Agree*

The workbook uses common, everyday language.

○ *Disagree*

○ *Agree*

Medical terms are used only to familiarize audience with the terms. When used, medical terms are defined.

○ *Disagree*

○ *Agree*

The workbook uses the active voice (e.g., Allison changed the flat tire versus the flat tire was changed by Allison).

○ *Disagree*

○ *Agree*

Numbers appearing in the workbook are clear and easy to understand.

○ *Disagree*

○ *Agree*

○ *N/A (No numbers)*

The workbook does not expect the user to perform calculations.

○ *Disagree*

○ *Agree*

The workbook breaks or "chunks" information into short sections.

○ *Disagree*

○ *Agree*

The workbook's sections have informative headers.

○ *Disagree*

○ *Agree*

The workbook presents information in a logical sequence.

○ *Disagree*

○ *Agree*

The workbook provides a summary.

○ *Disagree*

○ *Agree*

The workbook uses visual cues (e.g., arrows, boxes, bullets, bold, larger font, highlighting) to draw attention to key points.

○ *Disagree*

○ *Agree*

The workbook uses visual aids whenever they could make content more easily understood (e.g., illustration of healthy portion size).

○ *Disagree*

○ *Agree*

The workbook's visual aids reinforce rather than distract from the content.

○ *Disagree*

○ *Agree*

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○ *Agree*

The workbook uses illustrations and photographs that are clear and uncluttered.

○ *Disagree*

○ *Agree*

○ *N/A (no visual aids)*

The workbook uses simple tables with short and clear row and column headings.

○ *Disagree*

○ *Agree*

○ *N/A (no tables)*

Page Break

Section 2- Actionability

This section assesses whether materials are actionable and individuals of diverse backgrounds and varying levels of health literacy can identify what they need to do based on the information presented.

The workbook clearly identifies at least one action the user can take.

○ *Disagree*

○ *Agree*

The workbook addresses the user directly when describing actions.

○ *Disagree*

○ *Agree*

The workbook breaks down any action into manageable, explicit steps.

○ *Disagree*

○ *Agree*

The workbook provides a tangible tool (e.g., menu planners, checklists) whenever it could help the user take action.

○ *Disagree*

○ *Agree*

The workbook provides simple instructions or examples of how to perform calculations.

○ *Disagree*

○ *Agree*

○ *N/A (calculations)*

The workbook explains how to use the charts, graphs, tables, or diagrams to take actions.

○ *Disagree*

○ *Agree*

○ *N/A (No charts, graphs, tables, or diagrams)*

The workbook uses visual aids whenever they could make it easier to act on the instructions

○ *Disagree*

○ *Agree*

End of Block: PEMAT-P

Start of Block: satisfaction

How likely is it that you would use this workbook?

○ *Very Likely*

○ *Likely*

○ *Somewhat likely*

○ *Undecided*

○ *Somewhat Unlikely*

○ *Unlikely*

○ *Very Unlikely*

How likely would you be to recommend this workbook to an adolescent childhood cancer survivor who's about to transition into adult care?

- ○ *Very Likely*
- ○ *Likely*
- ○ *Somewhat likely*
- ○ *Undecided*
- ○ *Somewhat Unlikely*
- ○ *Unlikely*
- ○ *Very Unlikely*

Do you have any feedback for making this workbook more useful to survivors of childhood cancer or healthcare providers? If so, please use the space below to provide feedback.

____

____

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Do have any concerns about this workbook? If so, please use the space below to describe them.

If you have any additional feedback on the workbook, please provide it in the space below.

Thank you for your time!

Your contribution is much appreciated.

End of Block: satisfaction

Appendix F

Additional Ethics Documentation

Copy of Consent Form

Checklist

This checklist is to be completed and submitted with this consent form. It is to be removed from the final version of the consent document.

X Most recent version of consent template (November 2011) has been used
X Footer includes consent version, study name, line for patient initials
X Font size no less than 12 [except for footer]
X Left justification of text
X Grade 9 or lower reading level. Assessed reading level is: __8________
X Accepted definitions for specialized terms used where applicable
X Plain language principles used for study specific wording – no jargon, no acronyms, short words, short sentences, active voice and, where appropriate, bulleted lists

Standard, required wording (in bold type) has been used in the following sections:

	Yes	No
Introduction	X	☐
Benefits (Q6)	X	☐
Liability Statement (Q7)	X	☐
Privacy and confidentiality (Q8)	X	☐
Questions or problem (Q9)	X	☐
Signature page	X	☐
Signature page for minor/assenting participants if applicable	X	☐

If you have answered No to any of the above, please give the rationale for these changes below:

TCPS2 guidelines provide a list of the information required for informed consent. Please refer to TCPS2, Chapter 3, available at: http://www.pre.ethics.gc.ca/eng/policy-politique/initiatives/tcps2-eptc2/chapter3-chapitre3/.

The HREB Policy Manual provides detailed information on specific consent issues including: consent to research in emergency health situations; the use of substitute decision makers; assent for children; research involving special populations (children, cognitively impaired); managing consent in situations of difficult power relationships; and community consent to research involving Aboriginal communities. Please refer to the HREB Policy Manual on the HREA website: www.hrea.ca

Consent to Take Part in Research

TITLE: Improving the Transitions of Pediatric Cancer Survivors into Adult Care

INVESTIGATOR(S): Dr. Roger Chafe

You have been invited to take part in a research study. Taking part in this study is voluntary. It is up to you to decide whether to be in the study or not. You can decide not to take part in the study. If you decide to take part, you are free to leave at any time.

Before you decide, you need to understand what the study is for, what risks you might take and what benefits you might receive. This consent form explains the study.

Please read this carefully. Take as much time as you like. If you like, take it home to think about for a while. Mark anything you do not understand, or want explained better. After you have read it, please ask questions about anything that is not clear.

The researchers will:

- discuss the study with you
- answer your questions
- keep confidential any information which could identify you personally
- be available during the study to deal with problems and answer questions

1. Introduction/Background:

One of the most important health services issues for children and youth with chronic medical conditions is transitioning to adult care. Late adolescence can be a tumultuous period for people, yet it is during this period that patient's transition from the pediatric to the adult health care environment. As patients get closer to the age of majority, they are often expected to take more responsibility for managing their condition. The continuance of high-quality care for young adults requires that there are proper supports in place to bridge the transfer. Tragically, there is evidence that many patients with cancer do not have a smooth transition to adult care and have experienced negative outcomes. In Canada, we still do not even have an accurate picture of how these transfers are occurring for childhood cancer survivors who require continuance of care. Previous research in this area has mostly taken a disease-specific approach, focusing on issues related to a single condition.

2. Purpose of study:

Due to the improved outcomes of pediatric oncology patients, increasing numbers of survivors of childhood cancer are reaching adulthood and are transferred into adult care. Engaging a practical approach to attempt and improve these survivors' transition into adult care is a direct and concrete way of trying to address the emerging health care needs of this high risk group. This research project will help inform policies that will shape care pathways and transition programs in an area of cancer care for Newfoundlanders and Labradorians. The results of this research project may apply to the organization, administration, and provision of health care services across the province.

3. Description of the study procedures:

Our research team will employ a case study design, focusing on childhood cancer survivors youth transitioning to adult care. The case will focus on patient and provider experiences of transfers to adult care, available services to support transfers, needs and gaps in services, losses in access to services related to the move to adult care, barriers and facilitators of quality care through transition, innovative practices and suggestions for improvements. The cases will be based on document reviews and key informant interviews with pediatricians, adult physicians, nurses, health care managers and families who are involved in the transition to adult care.

4. Length of time:

Participants will be asked to participate in one interview lasting approximately 1 hour long.

5. Possible risks and discomforts:

There are no expected risks of participation in the study.

6. Benefits:

It is not known whether this study will benefit you directly, but it will hopefully help improve health service delivery for children and young adult patients in our province**.**

7. Liability statement:

Signing this form gives us your consent to be in this study. It tells us that you understand the information about the research study. When you sign this form, you do not give up your legal rights. Researchers or agencies involved in this research study still have their legal and professional responsibilities.

8. What about my privacy and confidentiality?

Protecting your privacy is an important part of this study. Every effort to protect your privacy will be made. However, it cannot be guaranteed. For example, we may be required by law to allow access to research records.

When you sign this consent form you give us permission to

- Collect information from you
- Share information with the people conducting the study
- Share information with the people responsible for protecting your safety

Access to records

The members of the research team will see study records that identify you by name.

Other people may need to look at the study records that identify you by name. This might include the research ethics board. You may ask to see the list of these people. They can look at your records only when supervised by a member of the research team.

Use of your study information

The research team will collect and use only the information they need for this research study.

This information will include [your job title or your relationship to individual with one of the conditions under investigation]. Your name and contact information will be kept secure by the research team. It will not be shared with others without your permission. Your name will not appear in any report or article published as a result of this study.

Information collected for this study will kept for five years.

If you decide to withdraw from the study, the information collected up to that time will continue to be used by the research team. It may not be removed. This information will only be used for the purposes of this study.

Information collected and used by the research team will be stored in a locked cabinet in Room 444 on a secure floor of the Janeway Hostel in the Janeway Pediatric Research Unit. Dr. Roger Chafe is the person responsible for keeping it secure.

Your access to records
You may ask Dr. Chafe to see the information that has been collected about you.

9. Questions or problems:

If you have any questions about taking part in this study, you can meet with the investigator who is in charge of the study at this institution. That person is: Dr. Roger Chafe.

Dr. Roger Chafe

Phone 777-2844

Roger.chafe@med.mun.ca

Or you can talk to someone who is not involved with the study at all, but can advise you on your rights as a participant in a research study. This person can be reached through:

Ethics Office

Health Research Ethics Authority

709-777-6974 or by email at info@hrea.ca

After signing this consent you will be given a copy.

Signature Page

Study title: Improving the Transitioning of Pediatric Patients with Chronic Conditions into Adult Care

Name of principal investigator: Dr. Roger Chafe

To be filled out and signed by the participant:

Please check as appropriate:

I have read the consent. Yes { } No { }

I have had the opportunity to ask questions/to discuss this study. Yes { } No { }

I have received satisfactory answers to all of my questions. Yes { } No { }

I have received enough information about the study. Yes { } No { }

I have spoken to Dr. Chafe and he has answered my questions Yes { } No { }

I understand that I am free to withdraw from the study:

- at any time
- without having to give a reason Yes { } No { }

I understand that it is my choice to be in the study and that I may not benefit. Yes { } No { }

I understand how my privacy is protected and my records kept confidential Yes { } No { }

I agree to be video/audio taped Yes { } No { }

I agree to take part in this study. Yes { } No { }

Signature of participant:________________________

Name printed: ________________________________

Year Month Day:______________________________

Signature of witness (if applicable):_______________

Name printed: ________________________________

Year Month Day:______________________________

To be signed by the investigator or person obtaining consent

I have explained this study to the best of my ability. I invited questions and gave answers. I believe that the participant fully understands what is involved in being in the study, any potential risks of the study and that he or she has freely chosen to be in the study.

Signature of investigator: _________________________

Name printed:__________________________________

Year Month Day:________________________________

Telephone number: ____________________________

SAMPLE SURVEY INVITATION EMAIL

Dear <Name>,

We invite you to participate in a research project entitled "Life After the Janeway"- A Transition Workbook for Childhood Cancer Survivors. This research project is focused on the transition from pediatric care to adult for childhood cancer survivors. We have created a workbook to help educate childhood cancer survivors about their health and the importance of aftercare. In this study we aim to assess the understandability and actionability as well as receive your overall feedback on the workbook. At the end of the study, we plan to share our findings with key stakeholders including the Janeway oncology program.

You have been invited to take part in this survey as you have been identified as <a key stakeholder, childhood cancer survivor, health care provider> who plays a role in helping provide opportunity for childhood cancer survivors to maximize their care. Should you wish to participate, the workbook can be found <here>. After reviewing the workbook in detail, please complete the following survey <link to survey>. This survey will take approximately 10 minutes to complete. This study is completely voluntary and your responses will be kept confidential. All your responses are anonymous and any demographic information collected will not be tied to any individual. Once you complete this survey, you cannot withdraw from the study. Data collected from survey will be kept secure in a password-protected computer for 5 years and then will be destroyed according to Memorial University policies.

If you have any questions about this survey, or difficulty in accessing the site or completing the survey, please contact Devonne Ryan at devonne.ryan@mun.ca or 709-728-1400. To speak with someone from the ethics office, who is not involved with the project please email info@hrea.ca or call 709-777-6974.

Thank you for taking the time out of your busy schedule to complete this survey. We look forward to your responses.

Sincerely,

Devonne Ryan