

# PAN-CANADIAN FRAMEWORK FOR CANCER SURVIVORSHIP RESEARCH





*Members of the Canadian Cancer Research Alliance are motivated by the belief that, through effective collaboration, Canadian cancer research funding organizations can maximize their collective impact on cancer control and accelerate discovery for the ultimate benefit of Canadians affected by cancer.*

PAN-CANADIAN  
FRAMEWORK FOR  
CANCER SURVIVORSHIP  
RESEARCH

MARCH 2017

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## MESSAGE FROM CCRA

From its inception, the Canadian Cancer Research Alliance (CCRA), an alliance of Canada's major cancer research funders, has worked to advance cancer research in Canada so that cancer patients will be the ultimate beneficiaries. *Target 2020*, formally articulates the Alliance's priorities to maximize the impact of shared targeted research investment and "patient experience" is one of the thematic areas identified in the strategy. This area is two-pronged, with one strategy focused on the development of a survivorship research framework and another focused on the development of a research framework for palliative and end-of-life care. This report is the former, striving to inform the cancer research funding community on how and what kinds of research are needed to support cancer survivorship research that will make a difference to cancer patients as they move from diagnosis to treatment and to the post-treatment phase.

The framework's development was informed by a strategic literature review, an analysis of the current state of cancer survivorship research funding, and data gathered from key informant interviews and an online survey of the broad stakeholder community. A working group composed of CCRA member representatives and an expert panel of survivorship researchers provided guidance and feedback to the consultant, Dr. Robin Urquhart and her team at the Nova Scotia Health Research Foundation,

as she synthesized the data and formulated recommendations within the context of a conceptual model.

It is our hope that this document provides CCRA members and other funders with critical direction on how to support, in collaborative way, cancer survivorship research. We look forward to working with members on implementing the recommendations contained within and on documenting our collective progress in this area.

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# EXECUTIVE SUMMARY

The impetus for this framework came from *Target 2020*, CCRA's current strategic plan, which lays out the need for a cancer survivorship research framework. To this end, this framework is intended to enable coordinated action among CCRA members, other funders, and key stakeholders with the goal to optimize the excellence, relevancy, and impact of cancer survivorship research in Canada. It is designed to help stakeholders conceptualize the many facets of survivorship research and assist in determining how to allocate research resources in a direct, effective, and efficient way. Cancer survivorship is a complex field, but also a relatively neglected phase of the cancer experience when compared to other phases of care.

Stakeholder input and views from across Canada were gathered through an online survey and key informant interviews, and was supplemented with findings from published studies and reports. In the context of the development work, a cancer survivor was defined as *a person who has had a cancer diagnosis, or has experienced cancer in some way*. Therefore, the term cancer survivor also included family/friend caregivers whose lives have been impacted by a cancer diagnosis. In addition, this framework considers the experiences of persons with metastatic disease, as there is a large population of people now living a long period of time after an advanced cancer diagnosis or recurrence.

To be effective, future research must respond to survivors' and health system's needs, build on existing strengths, avoid duplication of efforts, and have impact.

The four recommendations are:

1

### **ENSURE ONGOING AND MEANINGFUL INVOLVEMENT OF CANCER SURVIVORS AND THEIR FAMILY/FRIEND CAREGIVERS.**

- Involve survivors and their family/friend caregivers in setting research priorities and developing relevant and responsive funding calls.
- Strongly encourage the participation of survivors and their family/friend caregivers in the research process from the development of research questions to the interpretation and dissemination of research findings.

2

### **ALIGN FUNDING CALLS WITH EXISTING NEEDS AND POTENTIAL FOR IMPACT.**

- Invest in research priorities that address the gaps in cancer survivorship research and care that have been identified by stakeholders across Canada.
- Increase investment in intervention research, particularly interventions aimed at improving survivors' experiences and outcomes as well as preventing and managing the known late and long-term effects of cancer and its treatment.
- Invest in research that addresses the needs and outcomes of special populations.
- Invest in robust implementation science to identify, design, and test effective and efficient ways to move research findings into routine practice and policy.

The conceptual model for this framework is provided in Figure 1 on page 6.

## 3

### CREATE OPPORTUNITIES FOR THE TRANSLATION OF RESEARCH INTO PRACTICE AND POLICY.

- Actively support collaborative research that crosses jurisdictional, disciplinary, and professional boundaries.
- Enable health professional and decision-maker involvement in the design of funding calls to ensure relevant and responsive research is funded.
- Develop and implement a comprehensive evaluation framework to assess return on investment and promote successes in cancer survivorship research.

## 4

### BUILD AND MAINTAIN INFRASTRUCTURE AND EXPERTISE TO ADVANCE RESEARCH.

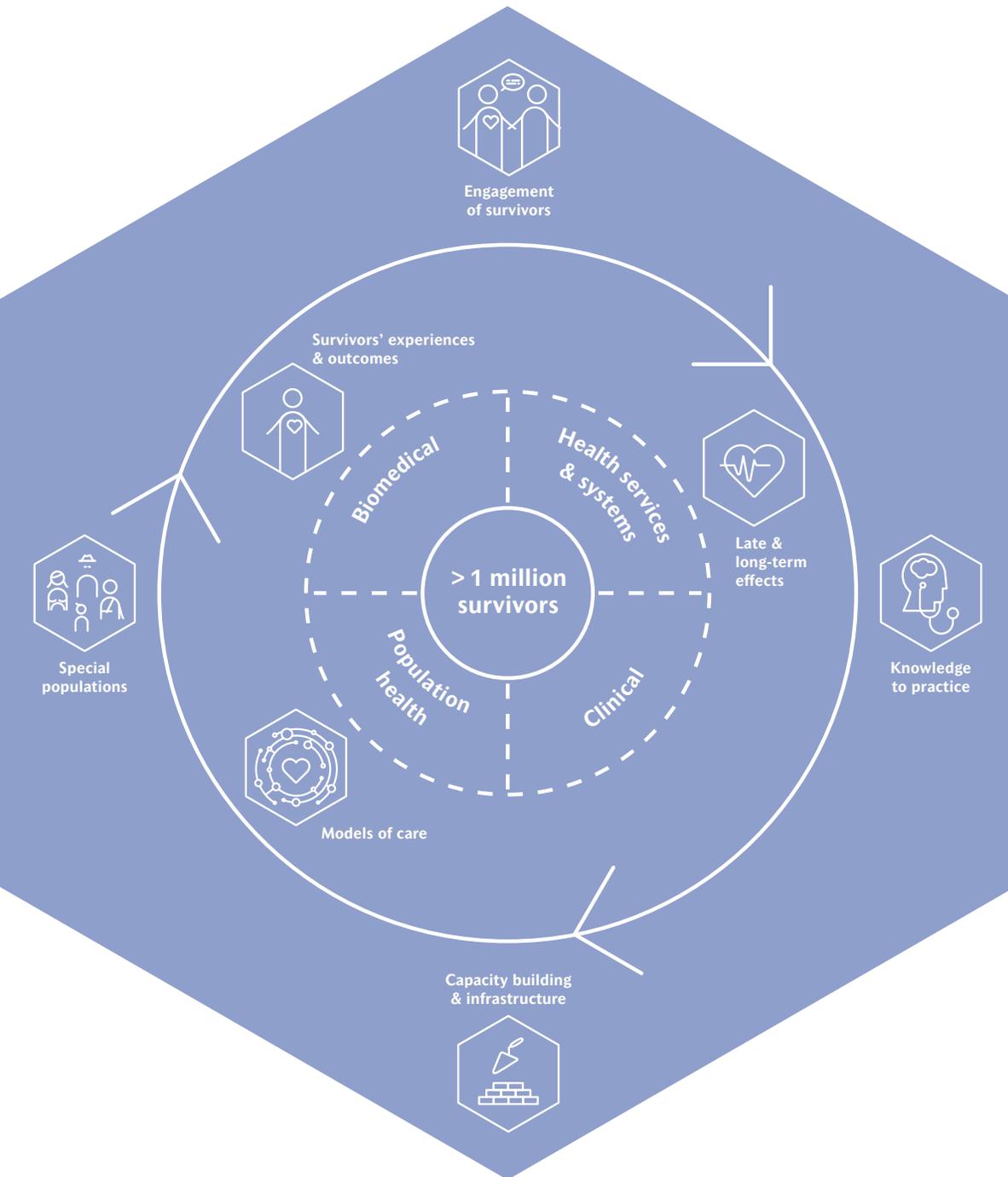
- Coordinate efforts to develop research tools and platforms that build on and leverage existing strengths in Canada that will have the greatest impact on the advancement of cancer survivorship research.
- Develop and sustain emphasis and expertise in cancer survivorship research.

The **Research Domains** encompass a range of content specific priorities to address key gaps in:

- Survivors' Experiences and Outcomes
- Late and Long-Term Effects
- Models of Care

In addition, **Cross-Cutting Themes** identify key issues that extend across all research domains regardless of content area or topic:

- Engagement of Survivors
- Special Populations
- Knowledge to Practice
- Capacity Building and Infrastructure



**FIGURE 1**  
**PAN-CANADIAN**  
**FRAMEWORK FOR CANCER**  
**SURVIVORSHIP RESEARCH**

**Research that is responsive to needs, builds on existing strengths, avoids duplication of efforts and has impact.**

- 1** Ensure ongoing and meaningful involvement of cancer survivors and their family/friend caregivers.
- 2** Align funding calls with existing needs and potential for impact.
- 3** Create opportunities for the translation of research into practice and policy.
- 4** Build and maintain infrastructure and expertise to advance research.

# 1. BACKGROUND

## 1.1 RATIONALE FOR A RESEARCH FRAMEWORK

Across all cancer sites, two-thirds of persons diagnosed today will be long-term survivors. That means the majority of cancer survivors will live more than five years after their diagnosis. In the 1940s, this number was a mere 25%. This phenomenal increase represents incredible success in cancer diagnosis, detection, and treatment. In fact, more than one million people in Canada today have had a cancer diagnosis. The number of people living with or beyond cancer will grow rapidly as the incidence of cancer increases with our aging population. We have all, in some way or another, been affected by cancer.

Despite this success, cancer and its treatment continue to have substantial and long-term impacts. After treatment, many survivors report medical and non-medical needs that are similar to those they face during treatment. These impacts and needs often vary depending on age, sex, gender, ethnicity, cancer site, and the type of treatment a person has received. Yet, compared to other phases of the cancer care experience, particularly diagnosis and treatment, the period after treatment has been largely neglected in advocacy, clinical practice, and research.

At the same time, the health system is challenged to deliver the most appropriate care to cancer survivors. This is, in part, due to concurrent demands on cancer specialist resources from increasing cancer incidence, new and more complex treatments, and insufficient human and material resources. Associated sectors, such as education and employment, are also challenged to provide appropriate and responsive programs and services for cancer survivors. In fact, we know there are economic consequences for persons who have had a cancer diagnosis. For example, research has demonstrated that medical costs and productivity losses are much higher for cancer survivors than for those without a cancer history.

Notwithstanding this context, Canada has major strengths in cancer survivorship research, with world-leading cancer survivorship researchers (in models of care, psychosocial oncology, and physical activity, among other areas) who have worked in this area for many years. Canada also has large, readily available population-based databases that can monitor cancer survivors' health care

utilization and outcomes over time and is internationally-renowned for advances in implementation science and cancer control. We can and must leverage these strengths to foster cutting-edge research that translates into widespread policy and practice changes to ensure that the research funded makes the greatest impact possible on the health and well-being of survivors.

## 1.2 DEFINING CANCER SURVIVORSHIP

Since the Institute of Medicine (IOM) published its seminal report *From Cancer Patient to Cancer Survivor: Lost in Transition* (Hewitt, Greenfield, & Stovall, 2006), cancer survivorship has been increasingly recognized as an important facet of the cancer experience. The U.S. National Cancer Institute's Office of Cancer Survivorship (OCS) considers any person diagnosed with cancer a survivor, from initial diagnosis until his or her death (President's Cancer Panel, 2004). The OCS also includes a survivor's family members, friends, and caregivers in its definition since these individuals are also affected by a cancer diagnosis. More commonly, however, survivorship has been defined as a distinct phase of the cancer trajectory that occurs between the completion of primary treatment (e.g., surgery, chemotherapy, and/or radiotherapy) and cancer recurrence or death (Hewitt et al., 2006; Rowland, 2006).

To develop this framework and obtain stakeholder input and views from across Canada, we defined a cancer survivor as *a person who has had a cancer diagnosis, or has experienced cancer in some way*. Therefore, in the context of this work the term cancer survivor includes survivors from the time of diagnosis and family/friend caregivers whose lives have been affected by the diagnosis. Though family/friend caregivers may have different experiences than persons diagnosed with cancer, they are nonetheless profoundly impacted by their loved one's diagnosis.

Furthermore, unlike the more common understandings of cancer survivorship, this framework includes persons with metastatic disease since many now live for long periods following an advanced cancer diagnosis or recurrence. However, it does not incorporate issues related to a person's end of life—for example, palliative care, end-of-life decision-making, and bereavement—since these issues

require a multi-dimensional research strategy of their own (Canadian Cancer Research Alliance, 2016). Through a broad definition, we were able to include all persons affected by cancer, and explore research strengths, gaps, and opportunities from multiple important perspectives.

In our external consultations, respondents nearly always described gaps related to this particular phase of the cancer trajectory. It is also noteworthy that this particular phase has been relatively overlooked in terms of research, advocacy, and clinical care, yet requires committed research funding if we are to ensure that cancer survivors live as healthy and active a life as possible for as long as possible.

While this framework largely addresses the post-primary treatment phase, it is important to recognize that what happens

before this time period will impact one's experience in the post-primary treatment phase of their journey. As one key informant highlighted, we must "leverage dialogues that begin at the commencement of diagnosis when decisions [are] being made that affect an individual's life going forward." For survivors, the cancer trajectory is not a siloed experience where the care they receive in one phase is isolated from all other phases; rather their experiences are often framed around the connectedness and continuity (or lack of it) of care in general as they move through various parts of the system (Easley et al., 2016). Thus, this framework does not negate research that addresses survivorship issues that may occur (or be mitigated) during the diagnostic and primary treatment phases of the cancer journey.

### 1.3 LEARNINGS FROM THE LITERATURE

The IOM's seminal report (Hewitt et al., 2006) was perhaps the first time that cancer survivorship became broadly recognized as a distinct phase of the cancer journey. The report made clear that many cancer survivors require ongoing medical care for years following their diagnosis and primary treatment. It also highlighted the significant challenges that many face as they transition back to their daily lives, with ongoing physical, psychosocial, and functional needs that often go unnoticed and therefore unaddressed. In fact, we know that cancer and its treatment can have substantial and long-term impacts on a survivor's health and quality of life, and that following treatment many survivors have continued medical and supportive care needs similar to during active treatment (Howell et al., 2011). These impacts and needs often vary depending on age, sex, gender, cancer site, treatment type, and other characteristics and circumstances. For example, pediatric, adolescent, and young adult cancer survivors often have different needs than survivors of adult-onset cancers (Grunfeld, Earle, & Stovall, 2011). These needs relate to issues as broad-ranging as changes in growth and development, schooling and education, work

and career development, building of social relationships, infertility, and transitioning from pediatric care to adult care.

In Canada, Miedema and colleagues have explored many of these needs in-depth through a series of studies investigating young adult cancer survivors' experiences (Easley, Miedema, & Robinson, 2013; Miedema, Easley, Robinson, & Jke, 2013; Miedema & Easley, 2012; Robinson, Miedema, & Easley, 2014). Their research has revealed a lack of age-specific follow-up care with inadequate information and support around issues such as fertility/infertility, sexuality, intimate relationships, and entry/re-entry into the educational system or workplace. In addition, their research with young breast cancer survivors has shown that barriers related to seeking and/or receiving survivorship care are often rooted in a complex set of gender roles as they navigate their lives as patients, mothers, workers, and caregivers. We also know that survivors of pediatric cancers are at significantly increased risk for major morbidity and premature mortality once they become adults (Hewitt et al., 2006; Hewitt et al., 2003; Hudson et al., 2003; Oeffinger & Hudson, 2004). In fact, the Childhood,

Adolescent, Young Adult Cancer Survivors (CAYACS) program in B.C. has generated much evidence around the excess morbidity and mortality in pediatric, adolescent, and young adult cancer survivors. Using population-based data, the research group has shown that long-term cancer survivors who were diagnosed before 20 years of age are at increased risk of all types of morbidity, except those related to pregnancy and birth complications, compared to matched peers without a cancer history (Lorenzi et al., 2011). Hospitalizations in pediatric, adolescent, and young adult cancer survivors also greatly exceed that of their matched counterparts (Bhuller et al., 2016; Bradley et al., 2010; Lorenzi et al., 2011). This group has also shown that pediatric cancer survivors have poorer educational outcomes than children without a cancer diagnosis, particularly for survivors of central nervous system tumours (Lorenzi et al., 2009). On the other end of the age continuum, co-morbidity rates increase with age, resulting in greater medical complexity and arguably greater need for improved coordination of care compared to those without comorbid conditions (Christensen et al., 2009; Søgaard, et al., 2013). At the same time, poorer access to cancer services is well documented among elderly populations (Butler et al., 2013; Du

& Goodwin, 2001; Hutchins et al., 1999). Thus, older populations will also have unique needs and challenges with respect to coordinated, integrated survivorship care.

Fortunately, due to advances in early detection and treatment, the majority of persons diagnosed with a cancer today will be long-term survivors. As a result, these individuals now live long enough to be at risk for and potentially develop late and long-term effects of their cancer and its treatment. Some of these effects may first appear during treatment, but persist in a long-term chronic manner, while others may first present months or even years after treatment. Examples of common long-term effects include fatigue, neuropathies, cognitive impairments, sexual difficulties, anxiety, and depression. Examples of late effects that may present after treatment include musculoskeletal problems, major organ dysfunction or failure (e.g., heart and lung problems), and second cancers (Grunfeld et al., 2011). Risk factors for late and long-term effects can vary by cancer site, type of treatment, age, genetics, as well as social and environmental factors that influence functioning (Stein, Syrjala, & Andrykowski, 2008). As our knowledge of these effects has emerged, researchers have developed and tested interventions to prevent

and manage them. Of note, exercise and physical activity interventions have been shown to be safe and effective at reducing late effects of cancer and/or its treatment, including anxiety, depression, and pain (Knobf, Musanti, & Dorward, 2007; Mishra et al., 2014). Despite growing knowledge in this area, there is much to understand with respect to risk factors, mechanisms, types, prevalence, and trajectories of late and long-term effects. Such information is paramount to developing more appropriate (risk-adapted) models to care for survivors during and after treatment and more effective interventions to prevent and ameliorate late and long-term effects.

Much research has found that the post-primary treatment phase of care represents a major time of angst for cancer survivors. In fact, survivors often describe this transition as one filled with distress, fear, and feelings of abandonment (Ganz et al., 2004; Jefford et al., 2008; Parry et al., 2011; Urquhart et al., 2012a). Many survivors report frequent guilt during this period, which influences their decisions not to seek care when experiencing late and long-term effects because they worry about taking cancer care providers' time away from patients with active cancer

(Roundtree, Giordano, Price, & Suarez-Almazor, 2011; Urquhart et al., 2012a).

A number of Canadian studies—conducted in different jurisdictions and cancer sites—have demonstrated that survivors lack access to timely information and supports with respect to survivorship issues, and often feel unprepared in regards to follow-up care and the long-term physical, psychological, and emotional effects of their cancer and its treatment (Easley et al., 2016; Jones et al., 2012; Sisler et al., 2012b; Urquhart et al., 2012a). Interventions that address and ameliorate the psychological, emotional, and informational needs of cancer survivors are key to improving survivors' health and well-being.

Given the increased awareness of many survivors' ongoing and substantive care needs, follow-up after primary treatment has become an established component of medical care. As the IOM has laid out, follow-up care for cancer survivors should include the following components: 1) cancer-related follow-up care to detect disease recurrence or new primary cancers and to prevent/manage late effects of treatment; 2) management of pre-existing chronic conditions; and 3) general preventative care (Hewitt et al., 2006). Traditionally, cancer survivors have

continued to receive care after their primary treatment from oncologists at specialist cancer clinics. From the health system perspective, however, we are challenged to provide appropriate follow-up care to cancer survivors, including coordination among providers (De et al., 2011; Grunfeld & Earle, 2010). Indeed, rising cancer incidence and more complex cancer treatments are occurring alongside an already constrained cancer system in terms of both human and material resources (Erikson, Salsberg, Forte, Bruinooge, & Goldstein, 2007; Grunfeld et al., 2000). Research has demonstrated that alternative models of cancer follow-up care are safe, effective, and cost-effective compared to the traditional practice of oncologist-led care (Grunfeld et al., 1996, 2006; Wattchow et al., 2006). Indeed, Howell and colleagues (2012) conducted a systematic review on models of care for adult cancer survivors and found that primary care- and nurse-led models are equivalent to oncologist-led care for colorectal and breast cancer survivors. Notably, *shared care* models have not been systematically operationalized or tested in adult cancer survivors (Howell et al., 2012). At the same time, there is substantial variation in survivorship care models and no consensus

on what constitutes a model of follow-up care (Halpern et al., 2015)—though all aim to improve the quality of survivors' care by delivering comprehensive, coordinated, and individualized follow-up care (Gilbert, Miller, Hollenbeck, Montie, & Wei, 2008).

In Canada, wide variation exists with respect to how and where we provide follow-up care (Grunfeld et al., 2006; Ristovski–Slijepcevic, 2008). There are many possible reasons for this, which have been highlighted in published literature. For example, cancer specialists often develop relationships with their patients during the treatment period, which can make it difficult to transfer responsibility for care back to primary care providers in the community (Audisio & Robertson, 2000; Kantsiper et al., 2009). For survivors, confusion over which provider is responsible for their cancer follow-up care (Cheung, Neville, Cameron, Cook, & Earle, 2009; Grunfeld, Hodgson, Giudice, & Moineddin, 2010; Miedema, Macdonald, & Tatemichi, 2003) and uncertainty about whether community-based providers have the expertise required to manage cancer follow-up (Kantsiper et al., 2009; Roundtree et al., 2011) may also contribute to the variation in follow-up practices. Moreover, when examining

follow-up care practices in Canadian jurisdictions, researchers have uncovered issues related to the quality of care including both under- and over-utilization of necessary tests and procedures (e.g., colonoscopy for colorectal cancer survivors) as well as use of tests and procedures with little to no benefit (e.g., bone scans for breast cancer survivors) (Grunfeld et al., 2010; Hodgson, Grunfeld, Gunraj, & Giudice, 2010; Sisler et al., 2012a; Urquhart et al., 2012b). Collectively, these issues all highlight the importance of information exchange between healthcare providers (informational continuity) and collaboration across healthcare sectors to organize care in a coherent and connected manner (management continuity) (Reid, McKendry, & Haggerty, 2002).

Recent systematic reviews from Canadian research teams have synthesized the evidence related to continuity and coordination of care for cancer survivors. A 2012 Cochrane systematic review found no effective interventions to improve continuity of care in the routine follow-up of cancer survivors that considered survivor, healthcare provider, and process outcomes (Aubin et al., 2012), underscoring the need for more investigation in this important area. A 2016 systematic review that evaluated models of care and/or interventions aimed at improving coordination between oncology and primary

care for persons with breast and/or colorectal cancer similarly found that the majority of studies showed no changes in patient, healthcare provider, or system outcomes (Tomasone et al., 2016). A complementary casebook project documented 24 on-the-ground Canadian initiatives designed to improve or support continuity and coordination of care between oncology and primary care, with 11 of these targeting the post-treatment phase of the continuum (Brouwers et al., 2016). Initiatives focused on the post-treatment phase ranged from transition care or well follow-up clinics, transition volunteer navigators, and disease pathways embedded into processes of care. The identification of effective initiatives, however, was precluded by the lack of evaluative data. Nevertheless, common implementation barriers and facilitators were identified across initiatives, including lack of care standardization (barrier) and the existence of clinical/program leaders and repurposing of existing resources (facilitators). Certainly, there is also a need to better understand the many factors, including organizational and system capacities that influence the adoption, implementation, and sustained use of models of care and other interventions that have been shown to be beneficial.

Though the volume of cancer survivorship research has grown considerably in the past decade, there is recognition that additional research is greatly needed if we are to achieve high-quality care in widespread, effective, and sustainable ways (McCabe et al., 2013). A recent American Society of Clinical Oncology survey identified a number of research gaps in cancer survivorship. These included a relative lack of research on: common cancers other than breast cancer, older survivors (65+ years); long-term survivors (>5 years post-diagnosis); interventions in young survivors (<21 years); biological mechanisms and genetic factors related to recurrence and late and long-term effects; and patterns and quality of survivorship care (Jacobsen et al., 2016). In a review of cancer survivorship research in the US and Europe, Rowland and colleagues (2013) highlighted a number of future research priorities, focused largely on the investigation of late and long-term effects and the development of risk-adapted models of follow-up care, as well as determining which countries are best suited (due to existing resources and expertise) to answer specific research questions. The latter recognizes the necessity to capitalize on existing strengths to most effectively and efficiently advance the field (Rowland et al., 2013).

## THE CANADIAN CONTEXT

The number of people surviving cancer has grown substantially due to important advances in early detection and treatment, with an estimated 3% of the population now cancer survivors. This means there are more than *one million Canadians* today who have had a cancer diagnosis. Given the dramatic improvements we have seen in survival rates in childhood cancers over the last two decades, more than 30,000 Canadians are now survivors of a childhood cancer. Despite these improvements, cancer survivors are at risk of a cancer recurrence and a new primary cancer. Many also live long enough to be at risk for late and long-term effects of their cancer and/or its treatments. While some survivors experience few late effects, others face substantial, debilitating, and life-threatening consequences. At the same time, our health systems have become challenged to meet the unique acute and long-term health needs of this growing population. As a result, cancer survivorship has become a strategic policy focus of cancer control organizations and cancer care programs across the country (BC Cancer Agency, 2016; Canadian Cancer Research Alliance, 2015; Canadian Partnership Against Cancer, 2012; Cancer Care Ontario, 2015).

From a research perspective, Canada has been a world leader in cancer survivorship on a number of fronts, including research on models of follow-up care, the impact of physical activity on survivors' outcomes, and interventions to address psychosocial needs. There have also been a number of key national initiatives and developments over the past decade to build research capacity, collaboration, and foci in cancer survivorship research. In 2008, the Canadian Partnership Against Cancer (CPAC) convened two workshops with invited participants from across Canada. The first workshop in March 2008 aimed to develop priorities for a Canadian cancer survivorship agenda. The aim of the second in November 2008 was to advance the cancer survivorship research agenda and, to this end, 30 unique research priorities identified. The top five priorities, which are encompassed in the research domains and cross-cutting themes of this framework, were: 1) measurement and development of appropriate tools for use in survivorship research; 2) determining optimal models of follow-up care; 3) effective interventions; 4) mechanisms underlying the long-term effects of a cancer diagnosis and its treatment; and 5) needs of unique populations (Kazanjian et al., 2010).

Furthermore, one of the recommendations from this latter workshop was to develop a national consortium to translate the priorities into an active and collaborative research agenda.

In 2010, another workshop was held, bringing together cancer survivorship researchers from across Canada, with the specific goal of developing a Canadian consortium for survivorship research. A detailed description of the rationale, activities, and outputs of this workshop have been published (Doll, Kazanjian, Smillie, Ward, & Chasen, 2012). This focused effort resulted in the Canadian Cancer Survivorship Research Consortium (<http://www.ccsrc.ca>), which works to build the infrastructure for national (and international) collaboration in cancer survivorship research. Specifically, the Consortium undertakes activities such as hosting national meetings for survivorship experts, bringing together researchers from across Canada to collaborate on competitive research funding proposals, and disseminating existing research to support its implementation in programs and policies.

Another pan-Canadian initiative is the establishment of the Canadian Centre for Applied Research in Cancer Control (<http://cc-arcc.ca>), of which the Canadian

Cancer Society is the founding funder. This research centre aims to improve cancer control and the delivery of cancer care through the conduct of interdisciplinary health services, economics, policy, and ethics research and coordinated knowledge translation. The centre also places large emphasis on capacity building through student education and training opportunities. Though not a cancer survivorship research centre per se, one of its five research themes is survivorship specifically as it relates to how health and social care systems can best address the physical, psychosocial, and economic impact of cancer on survivors and their families.

Alongside these national initiatives, there are numerous examples of targeted investment in cancer survivorship research. One such example is the ELLICSR Health, Wellness and Cancer Survivorship Centre at the University Health Network (<https://www.ellicsr.ca>), a \$3.7M infrastructure project funded largely by the Canada Foundation for Innovation and the Ontario Ministry of Research, Innovation and Science. ELLICSR is designed as a collaborative space for cancer survivorship research bringing together researchers, health professionals and cancer survivors.

Today, this 12,000 sq. ft. research facility houses an interdisciplinary research team and innovative self-management research programs with a focus on testing new approaches to predict, prevent and manage the late and long-term effects of cancer and its treatment, and evaluating innovative models of cancer care and support. Another example is the Canadian Institutes of Health Research's (CIHR) *Late Effects of Childhood Cancer Treatments Initiative*, which has funded four multidisciplinary teams to study why some children are more susceptible to late effects of treatment with the eventual goal of developing clinically relevant interventions to prevent and ameliorate the often debilitating late effects that many cancer survivors face. This initiative is an example of a strategic and coordinated partnership among several funders (i.e., CIHR, C<sup>17</sup> Council, Canadian Cancer Society, Cancer Research Society, Ontario Institute for Cancer Research, Pediatric Oncology Group of Ontario, and The Hospital for Sick Children) as well as with the Canadian Pediatric Cancer Genome Consortium. The CIHR's *Breast Cancer Initiative* is another example of targeted funding across numerous funders and relevant stakeholders to fund high priority survivorship research.

Despite this progress, there has been no integrated effort to date among research funders and other key stakeholders to put forward a Pan-Canadian cancer survivorship research agenda. Thus, research topics continue to be largely led by individual researchers/labs conducting a spectrum of research on survivorship. Yet, we know that the research we fund does not always make its way into practice and policy to the point where it benefits cancer survivors in a more lasting way. Right now in Canada there is tremendous opportunity to build on our momentum and successes, and optimize our capacity to conduct collaborative research that addresses the needs of cancer survivors, their family/friend caregivers, and our health systems. To do so, we must leverage our existing strengths and come together in a more coordinated, strategic fashion to enable world-leading, interdisciplinary research that informs best practices and leads to improved care and outcomes for cancer survivors.

## THE INTERNATIONAL CONTEXT

Alongside Canada, the US, UK, and Australia are key players in advancing cancer survivorship research globally. Shared priorities among all four countries include design and testing of effective care models and interventions, investigation of late and long-term effects of a cancer diagnosis and its treatment, and examination of the needs of special populations (Girgis & Butow, 2009). Importantly, research linkages exist across these four countries to collaborate on these priority issues in ways that can improve cancer survivors' experiences and outcomes in each country. For example, The Cancer & Primary Care Research Network has connected researchers across these countries and beyond to promote greater international collaboration in primary care and cancer research with a focus on follow-up care and survivorship.

The US has been the world leader in terms of major funding initiatives related to cancer survivorship. In 1996, the National Cancer Institute in the US established the Office of Cancer Survivorship (OCS)—in large part due to consumer advocacy—to champion and direct research that identifies and addresses the substantive challenges faced

by persons living long-term after a cancer diagnosis. Today, the OCS stimulates and funds research through a variety of funding mechanisms, and also connects researchers with health care providers and the public to build common and collaborative research agendas. Five subsequent reports from 2003 onward (Adler & Page, 2008; Centers for Disease Control and Prevention, 2004; Hewitt et al., 2006; Hewitt et al., 2003; President's Cancer Panel, 2004) brought national visibility and commitment to the substantive and long-term needs of cancer survivors and an obligation to invest in cancer survivorship research. In 2008, the National Cancer Survivorship Initiative was launched in the UK. Co-led by the Department of Health and Macmillan Cancer Support, its key aims are to understand the needs of survivors and develop the most effective models of services,

care, and support to address these needs (Jefford et al., 2012). Despite these initiatives, however, neither country has established a national framework to advance research on cancer survivorship.

European countries, other than the UK, have had less focus on cancer survivorship (Rowland et al., 2013), though the European Commission has recognized the importance of cancer survivorship in its 2009 launch of the European Partnership for Action Against Cancer (Commission of the European Communities, 2009). In 2011, numerous European oncology organizations and societies (e.g., European Organization for Research and Treatment of Cancer, European Society of Pediatric Oncology) presented key survivorship issues to the European Parliament to heighten awareness around the need to focus attention to this phase of the cancer control continuum.

## 1.4 THE CANADIAN CANCER SURVIVORSHIP RESEARCH LANDSCAPE

Canada's cancer research community is fortunate to have access to the data collected annually by CCRA that summarizes the cancer research landscape, including financial investments according to source of funding and area of research. This snapshot of activities enables CCRA members to identify gaps and opportunities for strategic investment and collaboration on an ongoing basis. The available data cover the years 2005 to 2013. Of note that there have been significant investments in cancer survivorship research in 2014 and these are not captured in these data. For more investment data, see Appendix A.

In terms of cancer survivorship research, the total investment increased from \$11 million in 2005 to \$19.7 million in 2013 and targeted investments rose over this time period, particularly in 2012 and 2013 (Figure 2). This increase mainly reflects the Canadian Cancer Society's support of the Canadian Centre for Applied Research in Cancer Control (through its survivorship stream) and the CIHR *Late Effects of Childhood Cancer Treatments Initiative*.

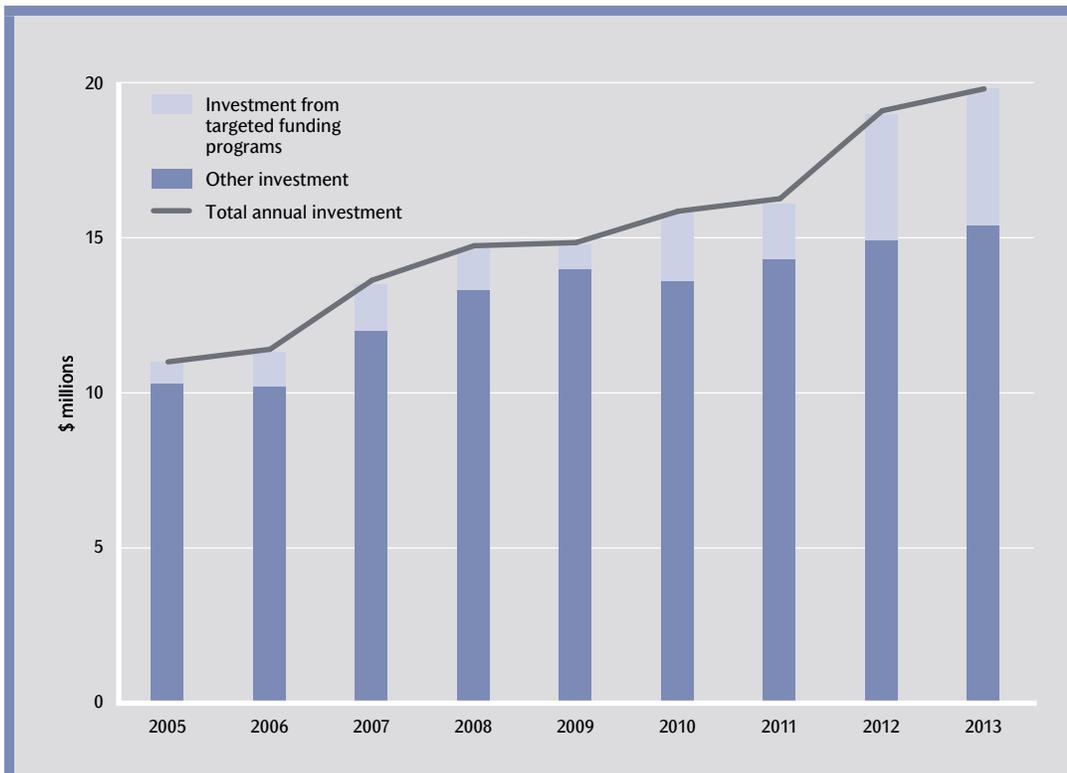
CIHR represented 42% and 39% of the total cancer survivorship research investments in 2005 and 2013, respectively, and the Canadian Cancer Society represented 23% and 17% of the investments in 2005 and 2013, respectively. Most of the increase occurred through operating grants. In fact, equipment and infrastructure grants, career awards, and trainee awards only saw small or modest changes. Of note, the increase in survivorship-related CIHR open operating grants from 2005 to 2013 was 151% (\$1.4 million in 2005; \$3.6 million in 2013).

The majority of the overall investment has been dedicated to specific cancer sites: 67% in 2005 and 63% in 2013. In addition, approximately one-quarter of the overall investment in both 2005 and 2013 went to breast cancer research (representing \$2.8 million in 2005 and \$5.3 million in 2013).

The physiological effects of cancer and its treatment have received substantially more funding than other research foci. In fact, 53% of all research funds invested in cancer survivorship research in 2013 went to investigating the physiological effects

of cancer and/or its treatment. In contrast, in the same year, 1.5% went to investigating social needs and supports and 2.5% went to investigating economic impacts, and these may represent gap areas in need of strategic funding.

**FIGURE 2**  
**INVESTMENT IN CANCER SURVIVORSHIP RESEARCH, 2005 TO 2013**



## 2. DEVELOPMENT OF THE FRAMEWORK

### 2.1 THE PROCESS

Oversight for this project was provided by a Working Group consisting of CCRA members and co-led by CPAC and the Alberta Cancer Foundation (see Acknowledgements). In addition, an Expert Panel was established, consisting of Canadian and international researchers with extensive expertise in survivorship research, decision-makers in cancer survivorship care/programs, and representatives with expertise in the health research funding landscape. The Expert Panel's role was to advise the Project Team on all aspects of framework development including pertinent

literature and reports, survey and key informant interview data tools, as well as the development of recommendations. Finally, an Advisory Panel was also established, consisting of representatives from community-based cancer organizations who provide services to and advocate for cancer survivors and family/friend caregivers, to provide the perspective of cancer survivors and family/friend caregivers throughout the project, and to assist the Project Team with engaging the cancer survivor community as needed.

*“Having common standards across the country, which CPAC facilitates, and having the weight of those recommendations is hugely helpful in selling it in the local environment. A Pan-Canadian strategy is very, very helpful and makes sense in our Canadian context.”*

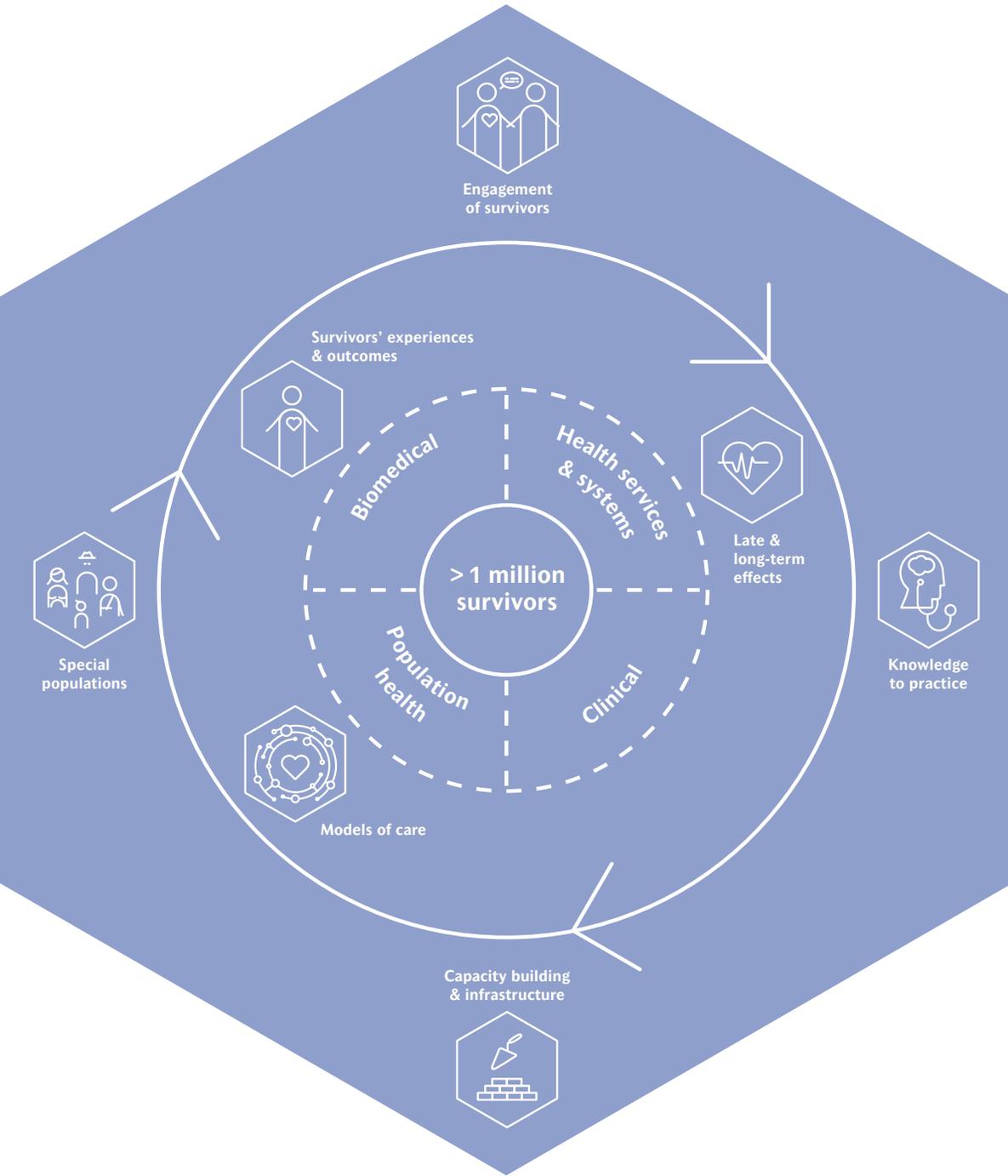
*– Decision-Maker*

The Project Team gathered data using various methods and from relevant stakeholder groups. Concurrent with scoping relevant published and grey literature, data collection involved surveys and key informant interviews to gather input from survivors, researchers, decision-makers and health professionals on critical areas of cancer survivorship research and how a research framework could address them. More than 200 stakeholders were consulted (see Appendix B).

In consultation with the CCRA Working Group and Expert Panel, common findings from the survey and interviews were organized into three overarching categories: Canadian Strengths, Research Domains, and Cross-Cutting Themes (see Figure 3). It was recognized, through survey responses and key informant interviews, that there are numerous existing strengths in cancer survivorship that should be leveraged to advance research in Canada. Respondents highlighted that Canada has world-leading researchers who conduct innovative and robust research across health pillars (e.g., biomedical, health services, population health, clinical) and have advanced

methodological expertise (e.g., database linkage, patient-reported outcomes, implementation science, qualitative). The population-based databases and registries, and associated infrastructure available across Canada are also major strengths that can and should be leveraged to track survivors' utilization of healthcare and long-term outcomes over time.

Furthermore, respondents indicated there is a wide range of potential opportunities available for trainees and new investigators that must be cultivated and supported. These include mentorship opportunities, which could be widely available due to the excellent clinical and research teams in place across the country with expertise in, or relevant skills that could be applied to, cancer survivorship research. Respondents suggest there is close communication between researchers with a willingness to work collaboratively and inter-professionally, fostering further opportunities for trainees and new investigators entering the field. With the growing resources and opportunities available, there are numerous strengths that can be leveraged to advance cancer survivorship research in Canada.



**FIGURE 3**  
**PAN-CANADIAN**  
**FRAMEWORK FOR CANCER**  
**SURVIVORSHIP RESEARCH**

**Research that is responsive to needs, builds on existing strengths, avoids duplication of efforts and has impact.**

- |   |  |   |   |
|---|--|---|---|
| <p><b>1</b><br/>                 Ensure ongoing and meaningful involvement of cancer survivors and their family/ friend caregivers.</p> | <p><b>2</b><br/>                 Align funding calls with existing needs and potential for impact.</p> | <p><b>3</b><br/>                 Create opportunities for the translation of research into practice and policy.</p> | <p><b>4</b><br/>                 Build and maintain infrastructure and expertise to advance research.</p> |
|---|--|---|---|

## 2.2 RECOMMENDATIONS AND PRIORITIES

The four recommendations are intended to enable coordinated action among CCRA members, other funders, and key stakeholders with the goal to optimize the excellence, relevancy, and impact of cancer survivorship research in Canada. Underlying these recommendations, it is imperative that future research responds to survivors' and health systems' needs, builds on existing strengths, avoids duplication of efforts, and has impact.

Within a coordinated and strategic approach, it is recognized that research is necessary across all four pillars of health research: biomedical, clinical, health services and systems, and population health. Despite organizing the key concepts in a structured and streamlined way, cancer survivorship is a complex field of research and each research domain and cross-cutting theme should not be considered a distinct entity—all components of the framework are interconnected and multifaceted.

## 1

**ENSURE ONGOING AND MEANINGFUL INVOLVEMENT OF CANCER SURVIVORS AND THEIR FAMILY/FRIEND CAREGIVERS.**

- Involve survivors and their family/friend caregivers in setting research priorities and developing relevant and responsive funding calls.
- Strongly encourage survivors' and their family/friend caregivers' participation in the research process from the development of research questions to the interpretation and dissemination of research findings.

## 2

**ALIGN FUNDING CALLS WITH EXISTING NEEDS AND POTENTIAL FOR IMPACT.**

- Invest in research priorities that address the gaps in cancer survivorship research and care that have been identified by stakeholders across Canada.
- Increase investment in intervention research, particularly interventions aimed at improving survivors' experiences and outcomes as well as preventing and managing the known late and long-term effects of cancer and its treatment.
- Invest in research that addresses the needs and outcomes of special populations. Key special populations include pediatrics, adolescents and young adults, Indigenous peoples, members of the LGBTQ community, rural and remote residents, and persons with metastatic and/or recurrent disease.
- Invest in robust implementation science to identify, design, and test effective and efficient ways to move research findings into routine practice and policy.

## 3

**CREATE OPPORTUNITIES FOR THE TRANSLATION OF RESEARCH INTO PRACTICE AND POLICY.**

- Actively support collaborative research that crosses jurisdictional, disciplinary, and professional boundaries. This is particularly important given cancer survivors' substantial and long-term needs, and the fact that survivorship care is resource intensive and crosses healthcare sectors.
- Enable health professional and decision-maker involvement (e.g., governments, health authorities, and accreditation bodies) in the design of funding calls to ensure relevant and responsive research is funded.
- Develop and implement a comprehensive evaluation framework to assess return on investment and promote successes in cancer survivorship research. This framework must include short- and long-term impacts at the survivor and health system levels, as well as be revised over time as needed.

## 4

**BUILD AND MAINTAIN INFRASTRUCTURE AND EXPERTISE TO ADVANCE RESEARCH.**

- Coordinate efforts to develop research tools and platforms that build on and leverage existing strengths in Canada that will have the greatest impact on the advancement of cancer survivorship research. One example could be the development of a comprehensive national longitudinal database to monitor survivor outcomes.
- Develop and sustain emphasis and expertise in cancer survivorship research. Strategic initiatives may include training and early career investigator awards, research chairs, team-based awards, or other funding calls to encourage early and mid-career researchers to apply their expertise to cancer survivorship research.

## 2.3 RESEARCH DOMAINS

The Research Domains outline broad research areas that encompass a range of content specific priorities. It is important to note these domains are not mutually exclusive—in the course of a survivor’s life and care, they are all interrelated.



### *Survivors’ Experiences & Outcomes*

This domain relates to survivors’ and their family/friend caregivers’ experiences and outcomes as they live with and beyond cancer. Issues within this domain focus on understanding their needs to enable optimal physical, psychological, social, emotional, and spiritual well-being. Psychological and emotional distress related to transitioning back to daily life and living with the possibility of a recurrence or second cancer are paramount to survivors’ experiences. This domain also incorporates the informational, functional, and economic impacts of cancer and its treatment, including survivors’ experiences both internal (e.g., navigation of the healthcare system) and external (e.g., work, social factors) to the healthcare system.

### **Stakeholder Perspectives**

Cancer survivorship research is rooted in providing the best outcomes for those who have experienced a cancer diagnosis and/or affected family/friend caregivers. The capacity to ensure the best possible outcomes depends on understanding the experience of cancer from survivors’ perspectives and the challenges they see as requiring attention. Survey respondents and key informant interviews continually emphasized the need for researchers to tackle the issues that most concern survivors themselves. Finding ways to optimize survivors’ physical, psychological, and emotional health are paramount. Even though family/friend caregivers’ have diverse experiences and are impacted quite differently compared to those diagnosed with cancer, their challenges and needs often fall under similar domains as persons who are diagnosed with cancer. Many who had experienced a cancer diagnosis and/or

their family/friend caregivers indicated that research that addresses their informational, psychosocial, and functional needs are key components of future work. However, due to the prominence placed on persons who have been diagnosed with cancer, the majority of gaps and challenges that surfaced from the survey respondents and key informant interviews focused on the experiences of survivors' post-treatment.

Many survivors felt unprepared for the period following primary treatment and reported high informational needs in this phase of their care. Informational needs span a wide range of issues including: risk of recurrence and new cancers; prevention and management of co-morbid conditions; evidence-based surveillance practices; and late and long-term effects of treatment. Regarding the latter, survivors and professionals (researchers, decision-makers, and health professionals) discussed how many survivors cope for months or years with fatigue and other chronic effects, which often come as a surprise, while others discussed adult survivors of pediatric cancers who are at risk for experiencing late and long-term effects of treatments that were completed decades ago. In effect, there was a strong perception that clear, consistent,

trusted, and timely information is a requisite to optimizing cancer survivors' experiences and outcomes. Survivors need to know what they can do to manage their health, what to watch out for, what to expect, and what to accept after completing their primary treatment. In addition, the development of strategies to inform and educate survivors and their current physicians of the risks and trajectories of late effects of treatments, particularly from treatments they received years ago, and appropriate treatment options were considered important.

Survey respondents repeatedly noted survivors' psychosocial burden is an area that requires a great deal of attention in order to improve the quality of life of survivors. It was generally acknowledged by survey respondents and interview informants that survivors have high needs with respect to psychosocial support and interventions are needed to address psychosocial issues. This was true throughout the cancer care experience, but particularly in the post-primary treatment phase when survivors are less intensively connected to oncology centres. Many expressed a sense of having been abandoned at the end of primary treatment. Psychosocial challenges include reintegration into social life under

the stigma of cancer, with survey and interview informants suggesting that this was something they and their friends, family, and co-workers often did not know how to talk about. As one survivor expressed in an interview, the feeling of isolation and difficulty connecting to peers' points to a need to look after "the whole person" and not just the biomedical elements of the disease. Survey respondents highlighted the need for better tools to cope with these challenges and made suggestions, such as professional psychological counseling or access to peer support services. However, the scale of unmet needs as indicated by the survey points to an opportunity for research to design and test innovative ways to provide support services. Creating new kinds of support and services, and modes of delivery, may succeed in reaching more survivors in a greater variety of circumstances, such as those uncomfortable with conventional psychological counseling or those in remote regions.

Many survivors expressed pervasive fear of cancer recurrence and a strong desire to find ways to improve their long-term survival. They expressed an interest in more research on lifestyle interventions and alternative therapies that would empower them to influence their outcomes,

particularly around preventative care and the management of existing chronic conditions. Such issues are indeed key components of optimal follow-up care (see Models of Care domain). Several survey respondents and key informants also suggested that lifestyle interventions, such as smoking cessation, dietary changes, and regular exercise that help survivors lower their risk of recurrence and improve their chances of long-term survival, need to be better understood and supported to encourage compliance. Interventions that take place outside of the health care system may be particularly challenging to design, test, and implement: for example, one professional informant noted the difficulty in obtaining funding and other support for exercise programs for survivors despite their proven benefits.

Another area of the survivor experience that survey respondents and key informants felt required concerted research attention relates to the economic and functional impacts of cancer, including the financial burden of cancer and return to work. In fact, survey respondents indicated these issues as prime concerns, with survivors placing a higher degree of priority on these issues than professionals (i.e., researchers, decision-makers and health professionals). Those without long-term disability insurance

or with expensive drug costs found themselves struggling financially. Likewise, several professional interview informants also expressed a need to use research to better respond to the economic impact of cancer on the survivor, family, and health system. One informant particularly called for research that addresses the needs of survivors in vulnerable socioeconomic circumstances. Survivors in such circumstances might not only live with additional stresses that come from financial insecurity, but struggle with caring for themselves post-treatment. Survey respondents noted that those struggling to return to work while also suffering fatigue and other long-term effects can find it difficult to afford medical and lifestyle interventions that could help them manage their health (e.g., medications, healthy food).

### **Priorities**

- Conduct research that targets the needs of survivors and assesses survivor-relevant outcomes, particularly for understudied cancer sites as well as for persons with metastatic disease.
- Investigate the functional and economic impacts and outcomes of cancer and its treatment.
- Investigate the informational and self-management needs of survivors with specific focus on navigating the healthcare system and managing comorbidities.
- Design and test interventions to address psychosocial needs, within specific focus on recurrence and transition back into daily life.



### *Late & Long-Term Effects*

This domain relates to the wide range of physical and psychological issues that may emerge during or after cancer treatment and persist in a chronic long-term manner. It also relates to problems that may not emerge until months or even years after a cancer diagnosis and its treatment. Examples include, but are certainly not limited to, fatigue, depression, infertility, learning and memory problems, musculoskeletal problems, heart problems, lung problems, and second cancers. This domain encompasses specific research areas such as the type, prevalence, trajectory, predictors, and mechanisms that underlie the development of late and long-term effects as well as the surveillance of survivors over time to identify the development or onset of late and long-term effects (e.g., pediatric cancer to adulthood) and targeted intervention research (lifestyle and medical) to prevent or mitigate the onset and development of late and long-term effects when sufficient evidence to design interventions exists. It includes the needs of patients coping with metastatic disease.

### **Stakeholder Perspectives**

Survey respondents and key informants strongly emphasized the need to better understand the long-term effects of surgery, chemotherapy, radiation, hormonal therapy, as well as support drugs like those that combat nausea. They were particularly concerned with morbidity and mortality related to major organ dysfunction, such as cardiac or lung problems, and second malignancies. They felt research is needed to determine who is at higher risk for developing such major late effects, and to ascertain what can be done to effectively identify and manage these as they emerge. This includes investigating genetic factors that increase the risk of late effects, such as cardiac failure. In addition, several survey respondents and key informants described a need to systematically collect information on late and long-term effects to better appreciate the risk and prevalence of these effects, particularly for specific sub-populations. Understanding the late and long-term effects of cancer and its treatment are of particular concern for pediatric patients who may have decades of survivorship post-treatment.

In the absence of important evidence on late and long-term effects and risks, follow-up guidelines are not able to provide comprehensive evidence-based recommendations for many important facets of survivorship care. Thus, one informant suggested that follow-up guidelines created by cancer care agencies err on the side of caution and recommend what may be inappropriately high levels of vigilance. Such hypervigilance is not without cost—excessive testing and reliance on specialists for unnecessary levels of follow-up care create an undue burden on the cancer system as well as unnecessary anxiety in survivors. Better information on what patient, tumour, and treatment characteristics are associated with elevated risks would allow for more appropriate surveillance and the development of risk-adapted models of long-term follow up care (see Models of Care domain) without undue risk as well as more efficient use of resources.

### **Priorities**

- Generate evidence to increase knowledge around types, prevalence, development, and trajectories of late and long-term effects.
- Investigate the mechanisms of late and long-term effects and predictors of high risk to inform treatment and follow-up decisions.
- Investigate the informational needs of patients and survivors related to late and long-term effects.
- Develop surveillance mechanisms to identify late effects and their impacts.
- Design and test interventions (lifestyle and medical) to prevent and ameliorate medical and psychosocial late effects.



### *Models of Care*

This domain relates to the organization and delivery of cancer survivorship care. It encompasses issues concerning continuity and integration of care, transitions in care, care settings, communication among providers and between survivors and providers, as well as the provision of follow-up care over time. Furthermore, Models of Care incorporates the need for implementation science to understand the contextual factors that influence care delivery as well as to ensure that evidence-informed models of care are put into practice and policy in more effective and efficient ways.

### **Stakeholder Perspectives**

The vast majority of survey respondents and key informants noted the importance of delivering optimal follow-up care to survivors, particularly in terms of addressing the IOM essential components for survivorship care: prevention and detection of disease recurrence, new primary cancers, and second cancers; prevention and management of the consequences of cancer and its treatment (e.g., late effects); and coordination among specialty and primary care to ensure all of the survivor's health needs are met, including pre-existing chronic conditions and general preventative care (Hewitt et al., 2006). At the same time, many also noted that determining the best way to organize and deliver this care remains challenging. Indeed, many survey respondents and interview informants raised concerns about the way that care is delivered in the post-treatment phase. These concerns often related to the care setting, transitions in care, communication issues, and the lack of appropriate, evidence-based surveillance practices.

Following treatment, many survivors continue to be under the active care of oncologists in specialist cancer centres when appropriate care could be delivered in non-specialist settings such as primary care. Respondents noted this is a situation that squanders the resources of specialists who could devote their energies more beneficially to newly diagnosed patients and those on primary treatment. According to one informant, “specialty clinics are clogged with patients who are being seen by some physicians who think it’s important to follow patients, often patients with a low risk of recurrence of their cancer, for [a] long period of time. This may be a real inappropriate use of resources with no significant benefit to the patient.” Needed research would seek to identify higher needs survivors and validate risk-adapted, evidence-based models of follow-up care. Such evidence would move us away from a ‘one-size-fits-all’ model of care to one that is informed by an individual’s needs, including the possible or probable consequences of his/her cancer and its treatments. One key informant also suggested the need to design and test “shared care” models that would include continued collaboration and coordination between primary and specialist care.

As a related issue, many survey respondents and key informants raised concerns about appropriate surveillance practices: namely, what tests and investigations need to be done, by whom, and when. For many cancers, evidence that would inform for appropriate follow-up care surveillance practices is lacking (see Late & Long-Term Effects domain). There was, however, recognition that surveillance practices will differ according to cancer type and treatment received, since these will impact survivors’ risks over the long term. It was also noted that the end of primary treatment does not coincide with the end of substantial and sometimes debilitating symptoms of the disease or effects of treatment. Once evidence for risk is established, it can address the question of when discharge from specialist care is appropriate, what tests and investigations are necessary (and when) for optimal surveillance, and the potential for overuse of resources for those who are not at risk of specific late effects.

Transitions in care, across the care continuum as well as across health care sectors, were seen as areas where intervention is needed. Respondents and informants highlighted the challenges in the transition of pediatric patients to adult care. One informant said bluntly, “I don’t think it goes very well. I think a lot of kids are lost to follow up, they don’t know – we don’t do it effectively so I think that’s a priority.” Many highlighted that communication is essential to facilitating optimal transitions in care, and perceived structural issues within our health care systems frequently impede good communication (for example, inadequate communication infrastructure across care settings and remuneration systems that do not always incentivize quality communication practices). Others also noted the transition to primary care presents challenges related to communication, survivors’ expectations, and provision of adequate psychosocial supports. Through the surveys, survivors expressed a common

fear that their primary care provider was not adequately informed about their follow-up care needs. Some expressed the need for improved training and preparation so that primary care providers can safely take on the responsibilities of follow-up care.

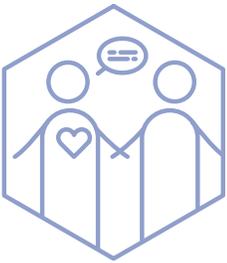
Finally, implementation science was identified as a research priority in this domain. This includes research to better understand the contextual factors that influence (and sometimes impede) the delivery of survivorship care in the primary care setting. In many cases, informants suggested the evidence to support changes exists, but there is a clear problem with finding successful ways to implement these changes in the real world.

**Priorities**

- Generate evidence to permit the development of risk-adapted long-term follow-up care, particularly for understudied cancer sites.
- Generate evidence to permit the identification of evidence-based surveillance practices as well as the identification of ineffective or low value tests, investigations and practices, particularly for understudied cancer sites.
- Design and test interventions to improve consistency, coordination and integration of follow-up care, including transitions in care.
- Conduct implementation science to understand how research findings move into routine practice and policy and how to remove ineffective or low-value practices from routine practice (i.e., de-adoption) and design more effective strategies to do both.

## 2.4 CROSS-CUTTING THEMES

The Cross-Cutting Themes recognize key issues that span across all research domains regardless of research area or topic, and outline priorities to address gaps that are prevalent across all research areas.



### *Engagement of Survivors*

This theme underlines the need for active and meaningful engagement of survivors and family/friend caregivers through a person-centered approach within both the research and decision-making processes. Survivors are the ultimate stakeholders in the research we fund and carry out. Thus, they and their family/friend caregivers should be active members of research teams to ensure relevant research questions and methods, and to support robust interpretation and communication of findings. This involvement may be enabled through numerous knowledge-to-action methods including, but not limited to, integrated knowledge translation, participatory-action research, and patient-oriented research. Aligning research with the needs and priorities of survivors and family/friend caregivers

will optimize its impact on real-world programs and policy. The voices of survivors themselves can act as key levers to ensure their issues are addressed by research and the resulting findings are translated at the point of care.

### **Stakeholder Perspectives**

Survey respondents and key informants emphasized a patient-centred approach to survivorship research is critical, and many perceived survivors should be engaged in research and policy/program decision-making processes in order to guide research and care priorities that best address their needs and concerns. From a research perspective, one way to accomplish this would be to include survivors and family/friend caregivers as full and active members of research teams. Multiple informants expressed views that

the typical engagement of survivors involves atypical representatives. That is, some felt that for a variety of reasons, survivors who are involved in research may not bring a fully representative range of viewpoints to the research process. For example, survivors involved in research may be more likely to resemble researchers in terms of education, income, and geographic location. As a result, these individuals felt researchers should look for ways to include a variety of patient voices as appropriate. One informant suggested researchers find ways to work with “vulnerable segments of the population, people with little education, immigrants or seniors. They are not the ones who are chosen...but they are probably the ones who would have the most to say on how our services should be organized.” One survivor informant suggested that survivorship forums might offer opportunities to engage with a variety of survivor voices beyond the individuals who typically agree to sit on committees/working groups or participate in surveys and interviews.

### **Priorities**

- Ensure engagement of survivors in both the research and program/policy decision-making processes.
- Leverage the voices of survivors to increase attention to and funding for cancer survivorship research.
- Gather perspectives and experiences from a broad range of survivors to support the generalizability of findings to more settings and populations.
- Design improved strategies and mechanisms for healthcare providers to communicate reliable information to survivors.



### *Special Populations*

This theme recognizes specific groups of survivors who may have distinct experiences and issues, depending on their characteristics and circumstances. Thus, focused research attention is warranted for these groups. Special populations include, but are not limited to, persons who have diverse cancer experiences based on sociodemographic factors (e.g., socioeconomic status, age, different cultural/ethnic backgrounds, sex, and gender), clinical differences (e.g., high-toxicity treatments, higher levels of comorbidity, long person-years of survivor life, and metastatic disease), system-based factors (e.g., those who are underserved due to lack of resources close to home), and other unique characteristics (e.g., those who identify as part of a marginalized population). Special populations also refer to survivors of understudied cancers, including, but not limited to, head and neck, brain, ovarian, and lung cancers.

### **Stakeholder Perspectives**

Although only five key informants identified as part of a special population group, gaps and priorities related to special populations were acknowledged by the majority of survey respondents and key informants as a critical theme to take into consideration across all cancer survivorship research domains. In fact, while responding to survey and interview questions, participants identified that survivors may have unique needs due to their sociodemographic characteristics, clinical and treatment history, or other personal characteristics. Participants also noted that system-based factors (e.g., a lack of resources close to home) may mean that certain groups of survivors require particular research attention. Survivors of understudied cancers may also represent special populations. The examples presented here represent the groups discussed by survey respondents and key informant interviews, but is by no means exhaustive. As several informants mentioned, appropriate and effective care and support considers the individual's circumstances, whatever they may be.

Survey respondents and key informants noted a number of sociodemographic characteristics that may result in distinct experiences and issues, including one's socioeconomic status, cultural/ethnic background, age, sex, and gender. For example, they highlighted a need to better understand how survivors' cultural and ethnic backgrounds impact their experiences and outcomes to tailor supports and interventions that meet their unique needs and contexts. One survivor emphasized there is a need for "research on different cultural needs, like ethnic cultural background—what a first generation Chinese woman would need versus what a rural Indigenous man would need." There was recognition that culture-specific responses and treatments are needed, particularly as they relate to members of Indigenous communities. For example, a Métis survivor noted the tendency to group all First Peoples under a single umbrella when it comes to research and care, which can be insensitive and counterproductive given the different cultures and circumstances of the peoples involved. A First Nations survivor identified a need to better understand the role and

effectiveness of traditional medicines. This respondent suggested that such medicines might have psychosocial benefits and that they will be used, regardless of whether there is established scientific merit in doing so. Furthermore, this respondent suggested that understanding potential interactions with conventional medical therapies is needed.

It was frequently noted that pediatric survivors have especially unique needs, particularly as they relate to late and long-term effects and to transitions of care as they move from pediatric to adult health care services. As one informant noted, pediatric survivors may have many decades of survivorship ahead of them and data related to long-term risks and appropriate surveillance mechanisms are particularly needed for this population.

Adolescent and young adult survivors were also identified by key informants as having many unique issues and needs. As one informant asserted, "Their problems are completely different than older people with cancer: fertility, young children, family, paying off a mortgage. All of that requires a lot of social work and psychological support." Another informant echoed similar

views, stating that the cancer system is set up to address where the cancer appears in the body and not where the patient is in his or her life. Yet the experience of cancer is significantly different for young adults than it is for the older adult population. Others noted that older adults have their own challenges as they may experience frailty or are more likely to experience comorbidities that present particular challenges for their care. Likewise, other characteristics and circumstances, such as whether a survivor is in a stable and supportive relationship or is single, is economically disadvantaged, or identifies with the LGBTQ community, have an impact on their supportive care needs, and these needs and how best to support these survivors require further research.

For survivors living in rural and remote areas, cancer survivorship often comes with challenges not faced by survivors living in more populous locations. One survivor repeatedly mentioned her isolation from both medical and psychosocial support following treatment. Seeing her treating oncologist required travel across a significant distance and even a primary physician was only available once a month. When discussing times when she had questions about her

physical health or stress and anxiety levels, she repeatedly said, “I had nowhere to go.” For survivors still undergoing treatment, the necessity of travelling long distances and staying away from their homes and communities for extended periods also creates psychological and financial strains that are often not addressed adequately by our health and social care systems. Another informant suggested a need to study the logistical problems and obstacles that treatment poses for patients, a particular concern for rural and remote residents who either travel significant distances for therapy while experiencing fatigue and other side effects, or who chose to live away from their homes, families, and community supports to receive treatment.

Additional factors were discussed in terms of elements and circumstances that can uniquely impact a survivor’s experience post-primary treatment. For example, the cancer site (i.e., brain, head and neck), types of treatment received, and metastatic disease can all create distinct survivorship needs. Of note, it was highlighted that survivors with metastatic disease often feel marginalized by the health care system, a circumstance that several survey respondents

called to be addressed. One respondent emphatically stated that metastatic survivors need more, not less, support than others. Another metastatic survivor commented in the survey, “I was told ‘there’s nothing more we can do for you.’ I felt that I had been set adrift...and needed to find my own support and information as to what happens next.” In summary, a range of factors can make each cancer experience unique and both survivors and professionals requested greater sensitivity to and research on how those factors influence survivors’ experiences and outcomes.

#### **Priority**

- Conduct research targeting understudied populations: these include, but are not limited to, specific cancer types, age groups, marginalized populations, and persons with metastatic disease.



### *Knowledge to Practice*

This theme pertains to the translation and application of research findings in real-world practice and policy to ensure we apply findings at the point where they actually benefit survivors and their family/friend caregivers. This includes communicating and tailoring information in understandable ways to appropriate audiences, and developing and using evidence-informed implementation and sustainability strategies that meet the needs of local settings. Collaborative research that includes true partnerships among stakeholders, including survivors, researchers, decision-makers, and health professionals, is key to finding innovative solutions that are more likely to have real-world impact.

### **Stakeholder Perspectives**

As many respondents of the professional survey and key informants suggested, research alone is not enough to positively impact the experiences and outcomes of cancer survivors. The findings must be applied to clinical practice, programs, and policy. Informants diverged in terms of what they felt were the barriers and solutions to moving research into practice. Many discussed the need for more targeted dissemination of what we already know. One informant suggested a need to collate comprehensive sets of best practices, especially for the most common forms of cancer, disseminate the recommendations, and identify those areas where gaps exist

so researchers can target those specific high need areas. Others suggested the problem lies less in synthesis and dissemination of existing research as much work has already been done in this realm (e.g., development of follow-up care guidelines) and more in the lack of active implementation of knowledge in a widespread and sustained way.

Multiple key informants commented on the obstacles that impede the implementation of research into practice. These include the busy pace of work in the clinical setting and the already constrained health human resources. One suggested that the amount of information available is too overwhelming for health professionals to “stay on top of it.” Another was particularly explicit in suggesting that physicians are overworked and their administrative environment is confusing. As a result, physicians cling to familiar routines and habits as a safety mechanism. Therefore, many felt the workload and environment of clinical settings need to be addressed before optimal translation of research into routine care can occur.

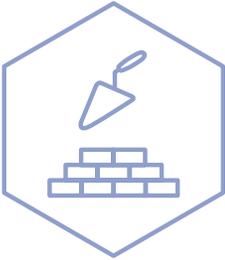
Many informants also identified obstacles that exist outside of the care setting. These include the cost of developing new programs (e.g., programs that could address late effects) and the lack of awareness on the part of decision-makers in regards to the very real and substantial needs of cancer survivors. Thus, many may be unwilling to commit funding and other resources to survivorship

programs that help individuals cope with a cancer diagnosis and manage their health post-primary treatment. As one informant suggested, "...patients who have a cancer diagnosis, I think to a large extent, have a post-traumatic stress syndrome after a diagnosis of cancer. This personal side is not well-appreciated. Physicians appreciate it, but policy-makers who are removed from clinical care may not realize the importance [of addressing the trauma]."

Numerous survey respondents and informants recognized that supporting and enabling interdisciplinary research teams that include researchers, decision-makers, health professionals, and survivor participation is one way to increase uptake of research into clinical settings and policy making. It was suggested that collaborative research would aid in increasing knowledge mobilization not only because health professionals and decision-makers will be familiar with the research, but also because the focus of the research would be more relevant to clinical needs. This notion is consistent with others who suggested that a comprehensive and justified plan for knowledge dissemination and translation must be a required component of research proposals. This would compel researchers to engage (on some level) with stakeholders from the beginning of the research process in order to ensure the research being pursued is useful and beneficial to the intended audiences.

### **Priorities**

- Prioritize knowledge translation, where appropriate, to ensure findings are communicated and delivered in understandable and relevant ways to stakeholders, including the public.
- Encourage collaboration across the research community, and with external stakeholders, to create interdisciplinary teams that include researchers, decision-makers, health professionals, and survivors.
- Provide funding and opportunities for researchers to share and translate their research into practice and policy.



### *Capacity Building & Infrastructure*

This theme relates to the strategies and mechanisms needed to further develop and advance research capacity and infrastructure to have the greatest impact for cancer survivors. It recognizes existing strengths within Canada and the necessity to capitalize on existing resources, data infrastructure, and expertise, and identifies priorities and opportunities related to research platforms and designs. The latter include developing tools and platforms relevant to cancer survivorship research (e.g., longitudinal cohort studies, survivor-specific instruments and tools), expanding our use of existing population-based data, and increasing capacity in intervention research. For example, strengthening infrastructure for longitudinal investigation would advance our understanding of cancer survivors' experiences and outcomes in the long-term, including our understanding of the prevalence and trajectory of late and long-term effects and the identification of still-unknown late effects. There is also a need to leverage the excellent mentorship from our world-leading researchers and support new researchers to focus their efforts and expertise across the breadth of cancer survivorship research.

### **Stakeholder Perspectives**

Survey respondents and key informants perceived an enormous need to build capacity and infrastructure in cancer survivorship research, primarily because this is a rapidly growing population that (unbeknownst to many) often experience substantive and sometimes debilitating effects long after primary treatment has ended. Encouraging trainees, early, and mid-career investigators to undertake research in cancer survivorship, supporting intervention research, and developing tools and platforms specifically for cancer survivorship research were among the suggestions made by respondents.

Informants typically agreed that grants targeting any aspect of survivorship would be valuable for increasing the number of trainees and new investigators, and thereby securing another generation of researchers working on cancer survivorship. One key informant noted that survivorship research (specifically referencing the post-primary treatment phase) does not have a high profile among students who prefer to work with patients in treatment and in oncology centres. Related, informants and survey respondents mentioned a need to more fully integrate aspects of cancer survivor care into medical school curricula. Another suggested developing three or four well-funded cancer survivorship centres across the country to provide key infrastructure for rigorous training in this area.

Survey respondents and key informants identified the need to invest in two types of study designs: intervention studies to improve survivors' experiences and outcomes and long-term cohort studies to gather data on the types, prevalence, and risks of late and long-term effects. To have high impact, such studies require collaboration across research teams and jurisdictions. The implementation of such studies would require longer term funding and coordination as well as an understanding of and willingness to fund and support projects that might not immediately generate benefits or results. One informant pointed to better coordination of funding among different agencies and jurisdictions as a way to address the large scale research needs. As he said, "I think we fund so many small scale studies that never amount to anything...these studies all float around out there in what we call, kind of, intermediate level science, you know, it's all suggestive, it's all a possibility. It all suggests it may be helpful and so on, but nobody does these large-scale, definitive trials that force policy makers to say if we're not giving these to cancer patients we are actually not following good science. But the studies that sort of move to that level have to be the big science studies, these large-scale studies that are done national level, sometimes even multi-national level."

Foundational elements for such "next level" research already exists in Canada. For example, survey respondents noted the presence of existing population-based databases and cancer registries that would be even more valuable if additional linkages could be accomplished. This includes linkages with clinical databases (e.g., laboratory and imaging databases) as well as across provinces. To fill knowledge gaps, systematic collection on rates of metastatic and recurrent disease is also warranted, which will require linkage with clinical data in the respective jurisdictions. Finally, there is growing expertise in Canada around patient-reported tools and outcomes. Leveraging this expertise to develop survivor-specific tools and instruments will advance research in this area.

**Priorities**

- Develop opportunities to build capacity in and increase support for trainees and new investigators.
- Increase capacity in intervention research, including cross-jurisdictional research that supports access to large and diverse populations.
- Leverage and build on existing resources, datasets and researchers to establish a strong research infrastructure.
- Develop tools and platforms for research that are relevant to cancer survivorship (e.g., longitudinal cohort studies, survivor-specific instruments and tools).

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## ABBREVIATIONS

<b>ARCC</b>	Canadian Centre for Applied Research in Cancer Control
<b>CAPCA</b>	Canadian Association of Provincial Cancer Agencies
<b>CAYACS</b>	Childhood, Adolescent, Young Adult Cancer Survivors
<b>CCAN</b>	Canadian Cancer Action Network
<b>CCRA</b>	Canadian Cancer Research Alliance
<b>CCSRC</b>	Canadian Cancer Survivorship Research Consortium
<b>CIHR</b>	Canadian Institutes of Health Research
<b>CPAC</b>	Canadian Partnership Against Cancer
<b>IOM</b>	Institute of Medicine (now known as the National Academy of Medicine)
<b>OCS</b>	Office of Cancer Survivorship (U.S. National Cancer Institute)
<b>YACC</b>	Young Adult Cancer Canada

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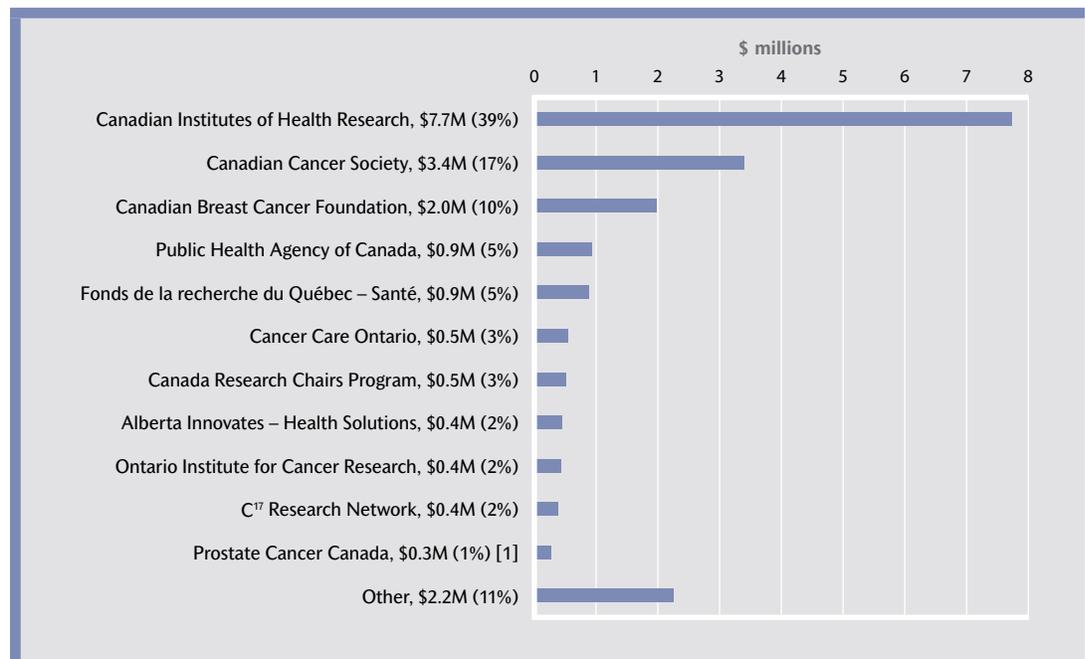
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## APPENDIX A. INVESTMENT IN CANCER SURVIVORSHIP RESEARCH

The following data come from CCRA's investment data on cancer research for the year 2013. The investment data come from a survey of 42 cancer research funders.

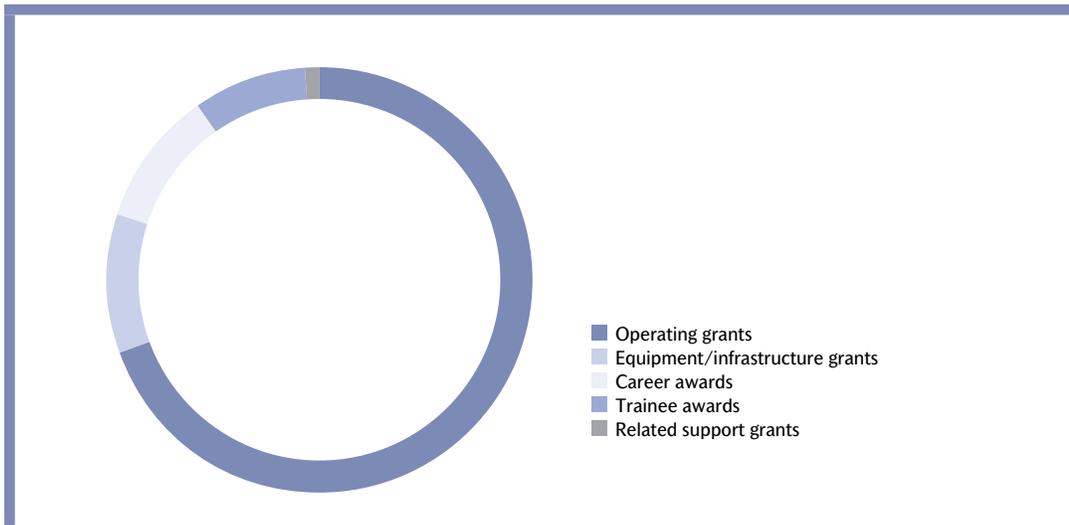
**FIGURE 4**  
**INVESTMENT IN CANCER SURVIVORSHIP RESEARCH IN CANADA BY FUNDER, 2013 (\$19.7M)**



[1] Prostate Cancer Canada's TrueNTH initiative, launched in 2014, has committed over \$8 million to 13 projects in 2014 and 2015.

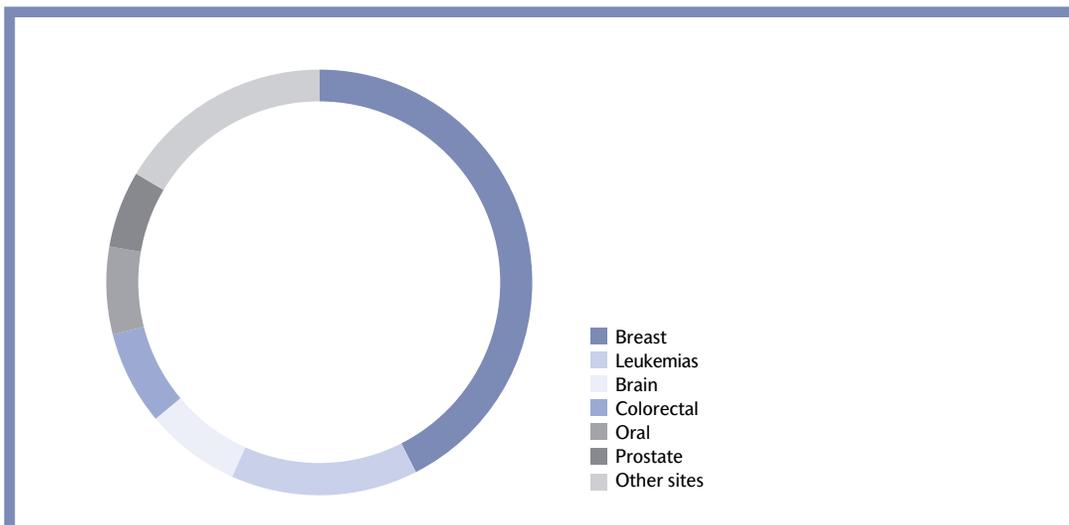
**FIGURE 5**

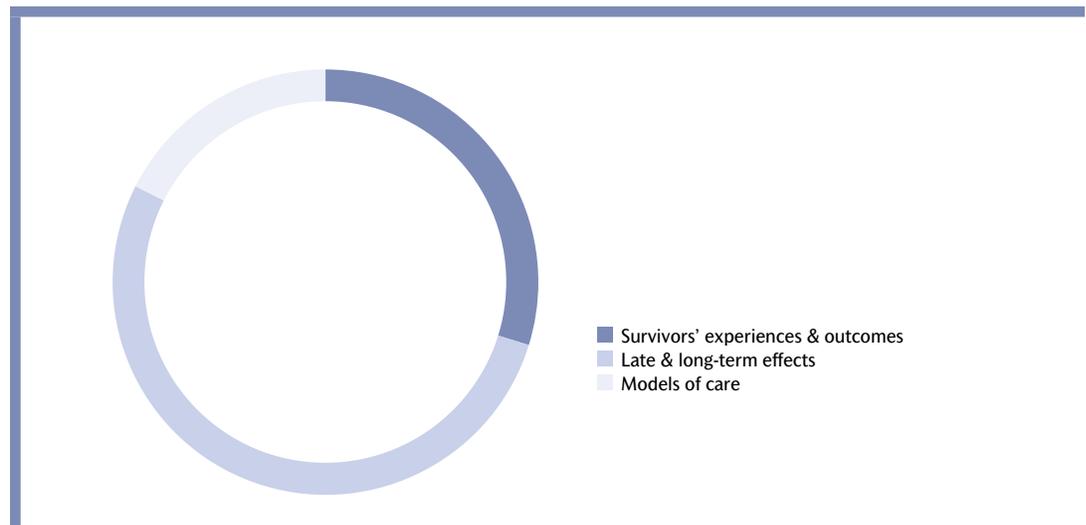
**DISTRIBUTION OF INVESTMENT IN CANCER SURVIVORSHIP RESEARCH IN CANADA BY FUNDING MECHANISM, 2013 (\$19.7M)**



**FIGURE 6**

**DISTRIBUTION OF INVESTMENT IN SITE-SPECIFIC CANCER SURVIVORSHIP RESEARCH IN CANADA BY CANCER SITE, 2013 (\$12.4M)**



**FIGURE 7****DISTRIBUTION OF INVESTMENT IN CANCER SURVIVORSHIP RESEARCH IN CANADA BY RESEARCH DOMAIN [1], 2013 (\$19.7M)**

[1] The research foci were grouped as best possible to the domains of the research framework as follows: Survivors' experiences & outcomes = Psychological effects, quality of life, social needs/support, economic sequelae, and thanatological issues; Late and long-term effects = Physiological effects; Models of care = Care delivery, access, and quality.

## APPENDIX B. EXTERNAL CONSULTATIONS

**In addition to a literature survey and consultations with working group members, external input was gathered through a series of key informant interviews and an online survey. Collectively, the input obtained from all these sources formed the basis for the development of the research framework.**

### KEY INFORMANT INTERVIEWS

Interview guides were constructed and refined in collaboration with the CCRA Working Group. Interview questions aimed to expand our understanding of the survey findings, including the gaps identified in the survey results. Questions were also included to gain further perspectives on the key ingredients of a successful framework, cross-cutting issues, and opportunities to move the framework forward. With assistance from the CCRA Working Group, an inclusive approach was taken when identifying key informants for our interviews. All informants were contacted by the Project Team via email, with reminder emails as required.

The interview guide was developed in two forms: one for survivors and one for professionals (researchers, decision-makers, and health professionals). Key informant interviews were conducted by telephone, audio recorded, and transcribed. Two members of the Project Team analyzed the transcripts using a thematic analysis approach (Guest et al., 2011). Nvivo Pro 11 (QSR International, Victoria, Australia) was used to organize and manage the data.

Sixteen key informant interviews were conducted with five survivors, nine professionals (researchers, decision-makers, and/or health professionals - many individuals held dual roles) and two international experts.

### Topics Covered in Interviews with Survivors

- Survivorship definition
- Areas for research
- Incorporating unique needs of survivors
- The role of survivors in the research process
- Key elements needed to build a successful research framework

### Topics Covered in Interviews with Professionals

- How their professional work relates to cancer survivorship.
- Survivorship definition
- Priority areas for research
- Incorporating unique needs of survivors
- Facilitators that help move survivorship research into practice and/or policy
- Key ingredients of a successful national research framework

- Recommendations with respect to strategically allocating research funds
- Challenges in building a survivorship research framework

Despite a diverse sample in terms of both participant characteristics and geographic location (see Table 1 below), similar themes (perspectives, gaps, and priorities) emerged across interviews with no new substantive information arising by the time of the final interview.

**TABLE 1**  
**DEMOGRAPHICS OF KEY INFORMANTS**

GROUP	N	GEOGRAPHIC LOCATION	N	SEX	N	SPECIAL POPULATIONS	N
• Cancer survivor	5	• Western Canada	5	• Female	7	• Pediatric/AYA Survivor	2
• Researcher	4	• Central Canada	4	• Male	9	• First Nations Survivor	1
• Decision maker	3	• Atlantic Canada	5			• Inuit Survivor	1
• Health professional		• International	2			• Métis Survivor	1
• Researcher & health professional	1						
• Researcher, decision maker & health professional	1						

## ONLINE SURVEY

Survey questions were constructed based on literature support and in collaboration with the CCRA Working Group, Expert Panel, and Advisory Panel. The survey was developed in two forms: one for survivors and one for professionals (researchers, decision-makers, and health professionals). Links to the surveys were distributed via email using multiple approaches. These included: a CCRA inventory of all funded researchers in cancer survivorship (many researchers were also health professionals in cancer care); a Project Team-generated list of key decision-makers in cancer survivorship care and programming in each province (e.g., relevant individuals from provincial cancer agencies or their equivalent, health authorities, and government); primary care and oncology network members, where they exist (e.g., B.C., Ontario); and email lists of survivor organizations and networks, including the Canadian Cancer Action Network (CCAN) and the organizations represented by members of the Advisory Panel. Participants were also asked to forward the link to other relevant individuals within their organizations or networks. A reminder email was sent to all the same individuals/networks approximately two weeks after the initial email invitation.

Data were collected through FluidSurveys. Responses to open-ended questions were qualitatively analyzed using a thematic analysis approach (Guest, MacQueen, & Namey, 2011). ATLAS.ti data software (Version 6.2, n.d.) was used to organize and manage the data. Three researchers from the Project Team analyzed the qualitative data to establish trustworthiness (Shenton, 2004).

## Questions for Both Survivors & Professionals

- Areas for new or further research: A. Care delivery and experiences; B. Physical symptoms and conditions; C. Psychological and social concerns; D. Quality of life; E. Health behaviours; F. Disease progression, recurrence, and survival; G. Research tools and platforms; H. Special populations; I. Economic impact of cancer
- Research areas with the potential to make the most impact to improve the lives and experiences of cancer survivors and their caregivers: after diagnosis; after completing primary treatment; long-term survivorship; recurrent disease/secondary cancer(s)/subsequent treatment(s); metastatic disease
- Greatest strengths of survivorship research happening in Canada

## Questions for Survivors

- Biggest gaps in cancer survivorship research in Canada

### Questions for Professionals

- Main barriers to implementing existing research evidence into survivorship care
- Biggest gaps in cancer survivorship research expertise
- Top three priorities for survivorship research
- Where Canada can make the biggest contribution/ impact to the global efforts in cancer survivorship research
- Views on specific elements of capacity: A. Do you feel there are sufficient numbers of trainees with content and methodological expertise to support and advance cancer survivorship research? B. Are there sufficient training and early investigator awards available to support capacity building in cancer survivorship research? C. Do you feel there is sufficient funding to support and advance the priority research areas you identified above? D. Do you feel there is sufficient infrastructure, resources, and research tools to support and advance cancer survivorship research? E. Do you feel institutions encourage collaboration in a way that supports and advances cancer survivorship research?
- Ranking of specific elements of capacity: expand the number of qualified trainees; increase support for new investigators; increase support for priority research areas; increase infrastructure, resources, and/or research tools; facilitate institutional collaboration

### Number of respondents

Of the surveys distributed, 185 participants completed the survey (114 survivors; 71 researchers, decision-makers, and health professionals).

### Demographics

In terms of 114 respondents to the survivor version of the survey:

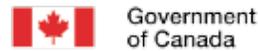
- 94 were survivors. For 61, the time since diagnosis had been six or more years.
- 65 were between the ages of 50 and 69
- 88 were female

Respondents reflected all regions in Canada as well as a mix of rural, mid-sized and large centres. The largest number of respondents indicated that they or their family/friend was a breast cancer survivor.

In terms of 71 respondents to the professional version of the survey:

- 53 were researchers
- 60 were between the ages of 35 and 69
- 47 were female
- 62 worked in Western or Central Canada
- 53 worked in communities of 500,000 or more

# OUR MEMBERS



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