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Models of Care that Include Primary Care for Adult Survivors of Childhood Cancer: A Realist Review

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Preface

The Agency for Healthcare Research and Quality (AHRQ), through its Evidence-based Practice Centers (EPCs), sponsors the development of evidence reports and technology assessments to assist public- and private-sector organizations in their efforts to improve the quality of healthcare in the United States.

The reports and assessments provide organizations with comprehensive, evidence-based information on common medical conditions and new healthcare technologies and strategies. They also identify research gaps in the selected scientific area, identify methodological and scientific weaknesses, suggest research needs, and move the field forward through an unbiased, evidence-based assessment of the available literature. The EPCs systematically review the relevant scientific literature on topics assigned to them by AHRQ and conduct additional analyses when appropriate prior to developing their reports and assessments. This report from the EPC Program at AHRQ is one of several efforts underway across the U.S. Department of Health and Human Services to implement provisions of the Childhood Cancer Survivorship, Treatment, Access, and Research (STAR) Act of 2018 (Public Law No: 115-180). The National Cancer Institute of the National Institutes of Health funded this report from the EPC Program at AHRQ.

To bring the broadest range of experts into the development of evidence reports and health technology assessments, AHRQ encourages the EPCs to form partnerships and enter into collaborations with other medical and research organizations. The EPCs work with these partner organizations to ensure that the evidence reports and technology assessments they produce will become building blocks for healthcare quality improvement projects throughout the Nation. The reports undergo peer review and public comment prior to their release as a final report.

AHRQ expects that the EPC evidence reports and technology assessments, when appropriate, will inform individual health plans, providers, and purchasers as well as the healthcare system as a whole by providing important information to help improve healthcare quality.

If you have comments on this evidence report, they may be sent by mail to the Task Order Officer named below at: Agency for Healthcare Research and Quality, 5600 Fishers Lane, Rockville, MD 20857, or by email to epc@ahrq.hhs.gov.

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Models of Care that Include Primary Care for Adult Survivors of Childhood Cancer

Structured Abstract

Objectives. We had two aims: (1) identify and analyze models of survivorship care for adult survivors of childhood cancer that include primary care, and (2) identify available tools, training, and survivorship resources for adult survivors of childhood cancer. For each aim, we used realist synthesis to provide insights on how and for whom, in what contexts, and via what mechanisms the models of care and resources we identified can be effective for adult survivors of childhood cancer.

Methods. We conducted a realist review. We developed an initial program theory through searches of the literature and discussions with Stakeholders. We then identified and summarized empiric evidence that supported or refuted the theory and developed specific hypotheses about how contexts and mechanisms may interact to produce outcomes (“CMO” hypotheses). The final program theory and CMO hypotheses were presented to Stakeholders for feedback.

Results. Our final refined theory describes how, within the overall environment, survivor and provider characteristics and facilitators/barriers interact to produce intermediate and final outcomes. We focus, in particular, on the role of models of care and resources in these interactions. From the theory, we developed seven CMO hypotheses (four focused on survivors and three focused on providers). The program theory variables seen most consistently in the literature include oncology versus primary care, survivor and provider knowledge, provider comfort treating childhood cancer survivors, communication and coordination between and among providers and survivors, and delivery/receipt of prevention and surveillance of late effects. In turn, these variables played the most prominent role in the CMO hypotheses.

Conclusions. To enable models of care that include primary care for adult survivors of childhood cancer, there needs to be communication of knowledge to both survivors and primary care providers. Our program theory provides guidance on the ways this knowledge could be shared, including the role of resources in doing so, and our CMO hypotheses suggest how the relationships illustrated in our theory could be associated with survivors living longer and feeling better through high-value care.

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Main Points

- Our program theory describes how survivor and provider characteristics and facilitators/barriers may interact to produce intermediate and final outcomes and the potential role of models and resources in these interactions.
- We developed seven hypotheses about the relationships between context, mechanism, and outcome (CMO) that could be associated with effective survivorship care models that include primary care.
- The program theory variables seen most consistently in the literature include oncology versus primary care, survivor and provider knowledge, provider comfort treating childhood cancer survivors, communication and coordination between and among providers and survivors, and delivery/receipt of prevention and surveillance of late effects. In turn, these variables played the most prominent role in the CMO hypotheses.
- Care delivered outside of the specialty setting needs to include communication of knowledge to both survivors and primary care providers; our program theory provides guidance on the ways this knowledge could be shared.

Background and Purpose

Childhood cancer survivors are at increased risk for life-long chronic morbidities owing to their cancer and its treatment. There is no consistent taxonomy for survivorship models of care. There is also a lack of clarity about which models are appropriate for whom and in what circumstances, as well as how resources (tools, training, resources, and processes) can support quality survivorship care.

The key decisional dilemmas for providers, survivors, caregivers, and health systems are (1) what models of childhood cancer survivorship care that include primary care may improve short- and long-term outcomes, for which survivors, and under what circumstances, and (2) what tools, trainings, resources, and processes can promote quality survivorship care across the various models?

Methods

We addressed contextual questions about the different types of models of care and resources through review of literature and discussions with Stakeholders. For the purposes of this project, the term “resources” includes long-term followup guidelines; educational materials; trainings; survivor care documents (e.g., survivorship care plans); survivorship care management processes (e.g., expedited routes of contact for consultation); and survivor supportive tools and services (e.g., support groups).

We addressed the decisional dilemmas by conducting a realist review. We interviewed Stakeholders (survivors, providers, researchers) and conducted targeted and iterative searching to (1) identify models of survivorship care and analyze the program theories (underlying ideas and assumptions) about how they are intended to work; and (2) identify available tools, training, and resources for childhood cancer survivorship care and analyze the program theories underlying

how they are intended to be implemented. Finally, we refined the identified program theory and developed hypotheses about context, mechanism, and outcomes through review of empiric evidence and discussions with Stakeholders.

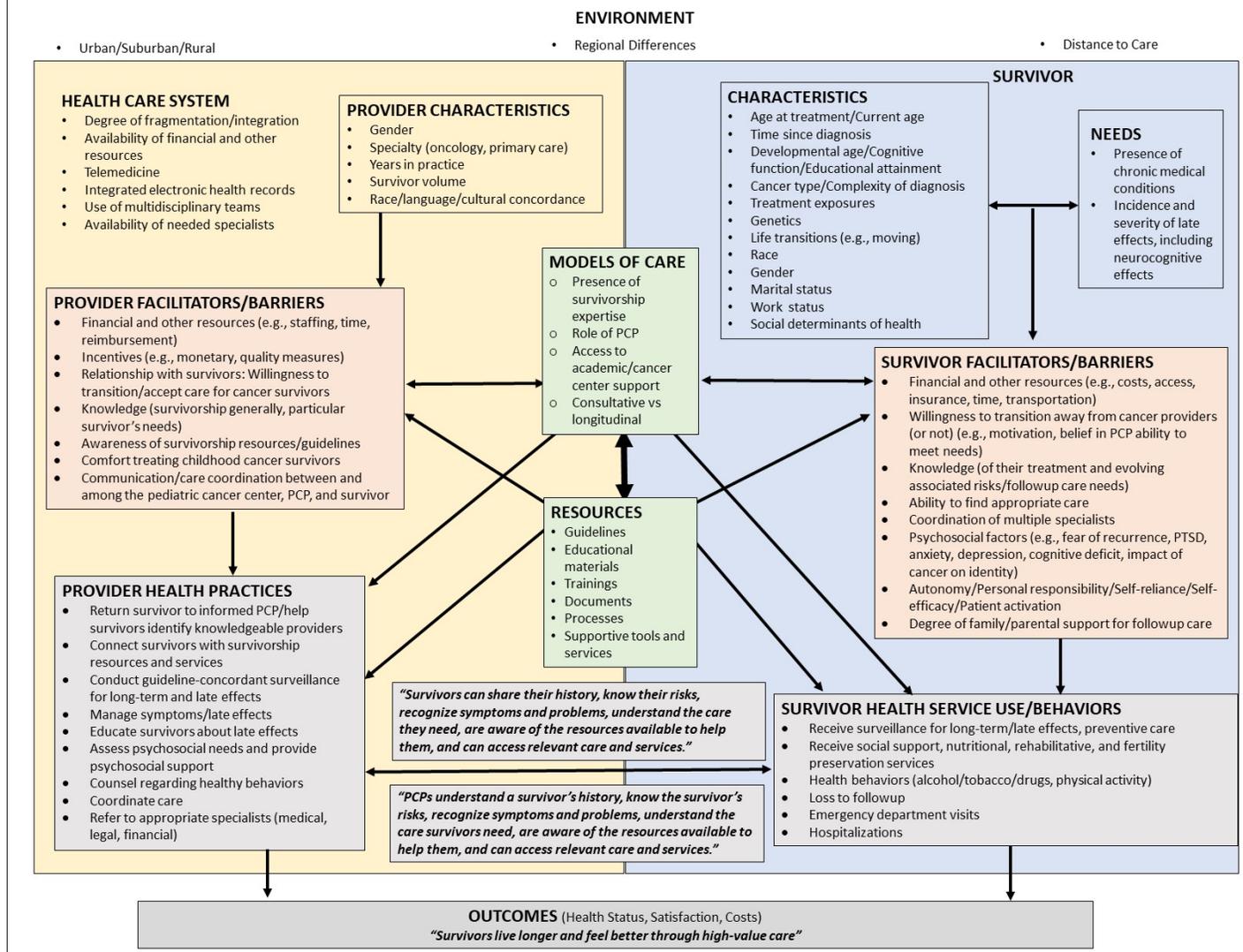
Results

There are an infinite number of models of survivorship care and there is no clear taxonomy for survivorship models of care. We identified four differentiating factors across models: (1) the inclusion of survivorship expertise (whether via a specialized primary care provider or oncologist; a physician, nurse practitioner/physician assistant, or multidisciplinary team); (2) the role of the primary care provider (e.g., main provider of survivorship care, provides survivorship care under the guidance of survivorship expert, provides primary care with no particular attention to survivorship); (3) degree of access to academic/cancer center support for survivors and/or providers; and (4) delivery of consultative versus longitudinal care. In practice, what is seen in the literature (and experienced by survivors) are more often patterns of care that occur not by design but owing to circumstance. We identified 41 resources freely available to both survivors and providers: 25 survivor-specific, 10 provider/researcher-specific, 6 for both. Discussions with Stakeholders suggested that resources are most helpful if they are easy to access, user-friendly, known to survivors and providers, from trustworthy sources, and valued by survivors and their families.

At the most basic level, the models of care that include primary care, and the resources, seek to provide information to survivors and/or primary care providers to enable them to obtain/deliver appropriate care. The program theory variables seen most consistently in the literature include oncology versus primary care, survivor and provider knowledge, provider comfort treating childhood cancer survivors, communication and coordination between and among providers and survivors, and delivery/receipt of prevention and surveillance of late effects. In turn, these variables played the most prominent role in hypothesizing what works for whom and in what circumstances. Our program theory illustrates how, within an environment, survivor and provider characteristics and facilitators/barriers, may connect through models of care and resources to achieve intermediate outcomes for survivors and providers, ultimately leading to survivors living longer and feeling better (Figure A).

We developed seven hypotheses about the relationships of context, mechanisms, and outcomes (Tables A and B).

Figure A. Refined Program Theory.



PCP = primary care provider; PTSD = post-traumatic stress disorder.

Table A. CMO hypotheses focusing on the survivor intermediate outcome.

In the CONTEXT of	this MECHANISM ... produces	OUTCOME (intermediate)	OUTCOME (final)
A) the availability of survivorship care plans, guidelines, and other resources	A1) improved survivor knowledge A2) information available to share with PCP to inform delivery of survivorship-related care	Survivors can share their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services.	Survivors live longer and feel better through high-value care.
B) healthier survivors (perceived or actual)	B) less perceived/actual need for survivorship-related care		
C) survivors engaged in health care system	C) improved knowledge		
D) survivor confidence in PCPs	D) willingness to transition care		

CMO = context-mechanism-outcome; PCP = primary care provider.

Table B. CMO hypotheses focusing on the provider intermediate outcome.

In the CONTEXT of	this MECHANISM ... produces	OUTCOME (intermediate)	OUTCOME (final)
A) the availability of survivorship care plans, guidelines, and other resources	A) information available to guide the PCP in delivering survivorship-related care	PCPs understand a survivor's history, know the survivor's risks, recognize symptoms and problems, understand the care survivors need, are aware of the resources available to help them, and can access relevant care and services.	Survivors live longer and feel better through high-value care.
B) shared-care with oncologist	B) support from the oncologist to aid the PCP in delivering survivorship-related care		
C) more experience caring for childhood cancer survivors	C) greater comfort caring for childhood cancer survivors		

CMO = context-mechanism-outcome; PCP = Primary Care Provider.

Limitations

Evidence on adult survivors of childhood cancer was limited leading us to consider studies of adult survivors of adult cancer. There was a lack of formal evaluations of models of care and data on final outcomes, particularly mortality, are sparse. A challenge was conducting a realist review of multiple ill-defined patterns of care rather than one intervention or model of care. Our review was also limited by the relatively short amount of time available, precluding the depth and number of iterative searches, syntheses, and refinement of theory typical in a realist review.

Implications and Conclusions

If care for adult survivors of childhood cancer is to be delivered outside of the specialty setting, there needs to be communication of knowledge to both survivors and primary care providers. Our program theory provides guidance on the ways this knowledge could be shared. Our context, mechanism, and outcome hypotheses suggest how the relationships illustrated in our theory could be associated with survivors living longer and feeling better through high-value care.

Introduction

Background

The American Cancer Society estimates that there were over 110,000 cancer survivors ages 0–19 as of January 1, 2019; when added to adult survivors of childhood (ages 0–14) and adolescent (ages 15–19) cancer, the total number of survivors approaches 400,000.¹ The growth in the number of survivors reflects new and more effective therapies, better risk stratification, and progress in supportive care that have improved treatment outcomes over the past decades, with 5-year relative survival increasing from 58 percent (1975–1977) to 84 percent (2008–2014) for children and from 68 percent (1975–1977) to 85 percent (2008–2014) for adolescents.¹ Unfortunately, owing to their cancer and its treatment, the majority of adult survivors experience life-long, chronic morbidities, such as cardiomyopathy, metabolic syndrome, and subsequent malignant neoplasms.²⁻⁶ However, the impacts of cancer and its treatment are not uniform, with different risks related to the specific type and location of previous cancer, its therapy, genetic predispositions, lifestyle behaviors, and comorbid health conditions.⁷ Thus, the National Academy of Medicine (NAM; formerly the Institute of Medicine) recommends life-long followup based on these factors.⁸

There is a lack of clarity regarding the appropriate models of survivorship care. Models of care range from specialized survivorship followup to general oncology followup to primary care followup. There are multiple barriers that preclude many childhood cancer survivors from receiving specialized long-term followup care, but there are also barriers that preclude those survivors from receiving quality survivorship care in primary care.^{9, 10}

Purpose and Scope of the Review

Given the growing number of childhood cancer survivors, the extensive morbidity and mortality experienced by these survivors, the need for risk-based survivorship care, and the lack of clarity regarding the appropriate models of childhood survivorship care, the key decisional dilemmas are (1) what models of childhood cancer survivorship care improve short- and long-term outcomes, for which survivors, and under what circumstances and (2) what tools, trainings, resources, and processes can promote quality survivorship care across the various models? The National Cancer Institute (NCI) requested a realist review as part of a series of projects for The Childhood Cancer Survivorship, Treatment, Access, and Research (STAR) Act to better understand the state of the science and ultimately improve the care and quality of life for childhood and adolescent cancer survivors.

We conducted a realist review to address two aims: (1) to identify and analyze models of survivorship care for adult survivors of childhood cancer that include primary care using realist synthesis and (2) to identify available tools, training, and survivorship resources for adult survivors of childhood cancer and use realist synthesis to understand how and why they produce effects on health outcomes.

Key Questions

Contextual Questions (CQ)

- CQ1. How is effectiveness defined and measured for survivorship care models for adult survivors of childhood cancer?
- CQ2. What are the models of survivorship care for adult survivors of childhood cancer?
- a. Which of these models include primary care?
 - i. What is the evidence of effectiveness of the different models that include primary care?
- CQ3. What survivorship care resources are available for adult survivors of childhood cancer and their families?
- a. What are the intended outcomes of the different resources available for adult survivors of childhood cancer and their families?
 - b. What is the evidence of effectiveness of the different resources available for adult survivors of childhood cancer and their families?
 - c. What are the monetary costs to access these resources?
- CQ4. What survivorship care resources are available to providers who care for adult survivors of childhood cancer?
- a. What are the intended outcomes of the different resources available to care providers?
 - b. What is the evidence of effectiveness of the different resources available to care providers?
 - c. What are the monetary costs to access these resources?

Key Questions (KQ) for the Realist Review

- KQ1. For whom and under what circumstances could different survivorship care models for adult survivors of childhood cancer (cancer diagnosed prior to age 21 years old) that include primary care be effective?
- a. What are the key mechanisms by which these models could be effective?
 - b. What are important contexts that determine whether different mechanisms could be effective?
- KQ2. For whom and under what circumstances could different survivorship care resources for adult survivors of childhood cancer be effective in achieving their intended outcomes?
- a. For survivors and their families
 - i. What are the key mechanisms by which these resources could lead to their intended outcome?

- ii. What are important contexts that determine whether different mechanisms could lead to outcomes?
- b. For care providers
 - i. What are the key mechanisms by which these resources could lead to their intended outcome?
 - ii. What are important contexts that determine whether different mechanisms could lead to outcomes?

For the purposes of this project, the term “resources” includes—

- long-term followup **guidelines**;
- **educational materials** directed at either survivor/family or care providers, regardless of media (i.e., electronic, hard copy);
- in-person or virtual **trainings** (i.e., workshops, conferences, continuing medical education courses) directed at either survivor/family or care providers;
- survivor care **documents** (i.e., survivor-specific standardized letters, treatment summaries, survivorship care plans);
- survivorship care management **processes** (i.e., expedited routes of contact for consultation, re-referral, support services; methods for digitizing and securely distributing health records; and other provider-to-provider and provider-to-survivor communications); and
- survivor **supportive tools and services** (in-person or digital), such as text messaging/peer navigator programs, support groups (in-person, telephone-based, or online), and professional psychosocial counselors (in-person, telephone-based, or online).

Methods

Rationale

A realist synthesis or review addresses the question “What works, how, why, for whom, to what extent and in what circumstances, in what respect and over what duration?”¹¹ In a realist review, the underlying ideas and assumptions about how an intervention works (known as program theories) are first identified and then tested and refined. Realist synthesis seeks to identify and understand how the mechanisms (M) through which an intervention or strategy works and its outcomes (O) are shaped by contextual features (C), such as the way the intervention is designed, how it is implemented and in what settings, and the different characteristics of recipients. We sought and synthesized empiric evidence to identify how different contexts trigger different mechanisms that result in different intended and unintended outcomes (CMO hypotheses).

We identified models of survivorship care and analyzed the program theories (underlying ideas and assumptions) about how they are intended to work. We identified available tools, training, and resources for childhood cancer survivorship care and analyzed the program theories underlying how they are intended to be implemented. And, finally, we tested and refined the identified program theories by reviewing empiric evidence evaluating the use of models and resources in practice to explain how different contexts shape the mechanisms through which they work.

Review Approach

Questions were initially identified by NCI and the Agency for Healthcare Quality and Research (AHRQ) and focused through a topic refinement process. We organized the questions as contextual questions (CQs) and realist review questions [or Key Questions, (KQs)]. Both the CQs and KQs were addressed through interviews with Stakeholders and literature review. The answers to the CQs describe the dimensions of models of care and resources for adult survivors of childhood cancer and their families and providers. The theory and CMO development process address the KQs by describing how models of care that include primary care and relevant resources could be effective. In practice, the theory and CMO hypotheses are the “answers” to the KQs.

Our protocol was posted for public comment by AHRQ, and we have followed the Realist And Meta-narrative Evidence Syntheses: Evolving Standards (RAMESES) guidelines in reporting our review.¹²

Stakeholder Engagement

We identified eight Stakeholders with a diverse range of perspectives, including clinicians with expertise in cancer survivorship and survivorship care, clinicians with expertise in cancer survivorship research, and childhood cancer survivorship patient advocates and caregivers. We convened small-group meetings of the Stakeholders at three stages throughout project to help clarify the relevant concepts related to models of care and resources (at theory development); to present an initial program theory and elicit feedback; and to elicit feedback on the refined program theory and CMO hypotheses. We engaged a realist synthesis expert throughout the project, as needed, to provide guidance on realist methods.

Development of Initial Program Theory

As with all realist reviews, our methods were iterative and included multiple searches of the literature and consultations with the Stakeholders. The objective was to develop an initial program theory and then refine it as our understanding grew based on the literature and our Stakeholders' input. For clarity, we used the term "model" to refer to "model of care" and "theory" to refer to program theory and mid-range theory.

We developed an initial program theory by engaging Stakeholders and reviewing mid-range theories and literature that described expectations for models of care and resources.^{13, 14} During this stage we sought evidence from a range of sources:

- Information from the team regarding their knowledge of mid-range theories related to access to care, knowledge specialization, coordination across the continuum of care, and uptake and use of resources.
- Opinion pieces, editorials, commentaries, and qualitative and mixed-methods studies about how models of care and resources are intended to work.
- Stakeholders' input on the initial program theory regarding how it did (or did not) reflect their experience and understanding of the issues, and on additional theories that may be useful for integration.¹⁵

Refinement of Initial Program Theory

Selection of Documents

We conducted additional searches to refine our theory by identifying empiric evidence that supported or refuted our initial program theory. This stage included—

- Searching three databases (PubMed, CINAHL, PsycInfo),
- Posting a notice seeking information on the federal register,¹⁶
- Conducting a general search of Google Scholar to identify documents relevant to 'childhood cancer' (or related terms) and survivorship,
- Using snowballing (searching references of studies) and berry picking (finding information bit by bit using a range of sources including expert input) to identify additional studies from key studies.^{15, 17, 18}

The initial program theory informed search terms for the databases. We did not limit our searches by study design or location of study. See Appendix A for the search strategy.

To screen abstracts and full-text articles, we followed methods described by Carrieri.¹⁹ Each abstract was screened by one of the team members. A random sample of the screened abstracts was checked by another team member for consistency.

Data Extraction and Appraisal

For program theory refinement, we focused on extracting evidence to support or refine fragments of the initial program theory or specific CMO hypotheses. We extracted study design and purpose; population of interest; and specific cohort, if applicable; as well as details on the models and/or resources under investigation. We identified the study variables, as defined in the initial program theory; variables with associations; and any other findings as they related to the initial program theory. We also drew from study authors' impressions to inform possible CMO

associations. Each team member completed data extraction; data extraction for each study was completed by one team member.

Included studies were classified as those making major, medium, or minor contributions in informing and testing the program theory.¹⁹ Our criteria for classifying studies were adapted from the categorization criteria from Carrieri et al., 2018:¹⁹

- Major: Evaluates a model of care or resource for adult survivors of childhood cancer
- Medium:
 - Provides insights regarding CMO connections in adult survivors of childhood or adult cancer and/or
 - Describes, but does not evaluate, a model of care or resource
- Minor: Conducted in non-cancer contexts but mechanisms could plausibly operate in the context of childhood cancer survivorship

Notably, we found so few studies classified as “major” that, in practice, we prioritized our evaluation of the literature by primarily drawing from the literature focused on childhood cancer survivors (both during childhood/adolescence/young adulthood and adulthood) and supplementing with evidence from adult survivors of adult cancers where relevant and informative.

Studies were also assessed as to whether (1) the data were relevant to the initial program theory (relevance) and (2) the research methods supported the conclusions (rigor). Determinations of rigor were informed by a set of questions derived from the Critical Appraisal Skills Programme (CASP) Checklist for Qualitative Research.²⁰

The following questions guided determination of rigor—

- Are the data credible and trustworthy?
- Are the methods appropriate to address the research goal?
- Was the research design justified?
- Was the data collected to address the research issue?

The following questions guided determination of relevance—

- Is the study population specific to adult survivors of childhood cancer?
- Is the setting within or outside of the U.S.?
- Are the findings generalizable to our program theory?

Analysis and Synthesis Process

We reviewed data extractions during team meetings. One team member summarized data extractions and grouped these by aspects of the program theory and CMO hypotheses. We reviewed findings to determine if they informed context, mechanism, or outcome, or associations between any two (C-M, C-O, M-O).²¹ Over a series of meetings, we discussed the implications of our findings for (1) refining the program theory, (2) updating the list of variables in the program theory, and (3) developing the CMO hypotheses.¹⁹

Final Program Theory

We conducted a final meeting with our Stakeholders during which we presented our revised program theory, including updates to the list of variables, and proposed CMO hypotheses. We elicited feedback regarding whether our findings reflected our Stakeholders’ understanding and experience. Based on this feedback, we made final revisions to the refined program theory, variables, and CMO hypotheses.

Peer Review

Experts in the fields of cancer survivorship, primary care, oncology, and realist review methodology, as well as caregivers, were invited to provide external peer review of the KQs and protocol prior to the review. AHRQ and representatives from NCI also provided comments. The draft report will be sent to peer reviewers and the Stakeholders, while simultaneously posted on the AHRQ website for 4 weeks for public comment.

Contextual Questions

The CQs were addressed by existing reviews and opinion pieces and through stakeholder input. A list of articles used to address the CQs can be found in Appendix B. The effectiveness questions for models and resources are addressed as part of the realist review (KQs 1 and 2) where we describe the evidence suggesting in what contexts and via what mechanisms models of care that include primary care and resources can be effective.

CQ 1: How is effectiveness defined and measured for survivorship care models for adult survivors of childhood cancer?

We identified, through Stakeholder engagement, a number of ways that effectiveness of survivorship care is measured:

- Loss to followup
- Surveillance for long-term and late effects
- Concordance with guidelines
- Communication, transfer of information, and care coordination
- Survivor returned to informed primary care providers/survivors able to identify knowledgeable providers
- Informed survivors who know what they need
- Survivors connected with needed resources and services
- Quality of life
- Morbidity
- Costs
- Mortality

CQ 2: What are the models of survivorship care for adult survivors of childhood cancer?

a. Which of these models include primary care?

i. What is the evidence of effectiveness of the different models that include primary care?

There is no consistent taxonomy for survivorship models of care, and models of care are rarely specifically selected in practice. The literature identified in this review generally provided evidence regarding who gets seen where and what care they receive (patterns of care) rather than formal evaluations of specific models of care.

The American Society of Clinical Oncology (ASCO) describes eight different models of care, classified primarily based on provider(s) and setting: oncology specialist care, multi-disciplinary survivorship clinics, disease/treatment-specific survivorship clinics, general survivorship clinics, consultative survivorship clinics, integrated survivorship clinics, community generalist model, and shared-care (Table 1).²² All eight of these models may incorporate PCP involvement. This listing is not specific to childhood cancer survivors, but it applies to this population.

However, studies generally have limited data on patterns of receipt of care for classification of models, and they tend to use broader categories. Using data from the Childhood Cancer Survivor Study, Nathan et al. organized receipt of health care as follows: no health care, general medical care, general survivor-focused care, and risk-based survivor-focused care.⁷ Oeffinger et

Table 1. Eight Models of Cancer Survivorship Care as Described by ASCO.

Model	Description
Oncology Specialist Care	<ul style="list-style-type: none"> • Follow up care occurs in the oncology setting with treating oncologist • Can be implemented in private practice, community hospitals or cancer centers • Can be implemented in disease-specific clinics or modality-specific practices • Communication and coordination with PCP*
Multi-Disciplinary Survivorship Clinic	<ul style="list-style-type: none"> • Care provided by a specialized team (e.g., oncologist, psychologist, cardiologist) in a separate clinical area • Can be implemented at cancer centers and other facilities with extensive resources • Ideal patient populations include: pediatric cancer survivors, AYA survivors and adult survivors of pediatric cancer[†] • Communication and coordination with PCP*
Disease/Treatment-Specific Survivor Clinic	<ul style="list-style-type: none"> • Care can be provided by a physician, NP, PA, or multispecialty team • Care provided in oncology setting • Can be developed for a common diagnosis, such as breast cancer, or treatment modality, such as stem cell transplant • Can be developed in private practice, community hospital, or cancer center • Communication and coordination with PCP*
General Survivorship Clinic	<ul style="list-style-type: none"> • Care can be provided by MD, NP, or PA (not multispecialty) • Can be implemented at a cancer center, community hospital, or private practice • Communication and coordination with PCP*
Consultative Survivorship Clinic	<ul style="list-style-type: none"> • Care can be provided by MD, NP, or PA • One-time visit with no follow up, but has potential for another visit • Ideal for providing treatment summary and care plan • Can be implemented at a cancer center, community hospital, or private practice • Communication and coordination with PCP *
Integrated Survivorship Clinic	<ul style="list-style-type: none"> • Embedded in the treatment focused oncology setting • Care can be provided by an MD, NP, or PA • Care is ongoing • Can be implemented at a cancer center, community hospital, or private practice • Provider communicates and coordinates care with PCP and specialists as needed*
Community Generalist Model	<ul style="list-style-type: none"> • Survivorship care is provided by PCP, NP, or PA* • Care setting can be at a health care system or private practice • Communication and coordination with PCP*
Shared-Care of Survivor	<ul style="list-style-type: none"> • Care for all survivors is coordinated between oncology specialist, and PCP generalists* <ul style="list-style-type: none"> <i>Without Transition</i> <ul style="list-style-type: none"> • Follow up care occurs in oncology setting • Can be implemented at a cancer center, community hospital, or private practice <i>With Transition</i> <ul style="list-style-type: none"> • Consultation with the oncology specialists occurs as needed • At a predetermined time, care is transitioned to the PCP only*

* primary care provider (PCP) relevant characteristic(s)

[†] related to childhood cancer survivors

ASCO = American Society of Clinical Oncology; AYA = adolescent/young adult; MD = medical doctor; NP = nurse practitioner; PA = physician's assistant; PCP = primary care provider.

al. focused on health care system interactions: general contact, general physical examination, cancer-related medical visit, and cancer center visit.²³ Mueller et al. organized care by provider type seen: primary care provider (PCP), specialty care physician, nurse practitioner (NP)/physician's assistant (PA), and survivorship clinic team.²⁴ Surveys of providers have asked about categories such as continued care in pediatric oncology, referral to primary care, shared-care with primary care, and followup through a specialized long-term followup program.²⁵⁻²⁷ One systematic review simply categorized models as general practitioner (GP) led versus shared care between a GP and pediatric oncology or a late-effects clinic.²⁸

In discussions, Stakeholders described several aspects defining different models of care, including:

- Longitudinal versus one-time clinics
- Location where care is delivered (e.g., cancer center clinics vs. community care)
- Who delivers the care (e.g., doctor, advance practice provider, nurse, or combination thereof)
- Degree of specialized survivorship knowledge – applies to both PCPs and oncologists
- Role of PCPs who specialize in cancers survivors and may be integrated in a cancer center, located at a community hospital, or in a private practice
 - Either solo or part of an internal medicine-based survivorship clinic

Using the information from the literature and discussion with Stakeholders, we defined four dimensions describing models of survivorship care:

- Particular expertise in survivorship? [yes/no/unclear]
 - Is the survivorship expert trained in oncology, primary care, other, unclear?
 - Is the survivorship expertise MD, NP/PA, multidisciplinary, unclear?
- Role of PCP [main provider of survivorship care, provides survivorship care under the guidance of survivorship expert, provides primary care with no particular attention to survivorship, unclear]
- Access to academic/cancer center support for survivors and/or providers [yes/no/unclear]
- Consultative versus longitudinal or unclear

CQs 3 and 4. What survivorship care resources are available for adult survivors of childhood cancer and their families or to providers who care for adult survivors of childhood cancer?

- a. What are the intended outcomes of the different resources available for adult survivors of childhood cancer and their families or their care providers?
- b. What is the evidence of effectiveness of the different resources available for adult survivors of childhood cancer and their families or their care providers?
- c. What are the monetary costs to access these resources?

In contrast to the taxonomy for the models of survivorship care, the taxonomy for the tools/training/resources/processes (hereinafter “resources”) is more straightforward. Surveys conducted by Nathan et al. and Suh et al. have asked about the value of options such as survivor-specific standardized letters; long-term followup guidelines; expedited routes of contact for

consultation, re-referral, and support services; and websites, medical education, and pamphlets.²⁶ ²⁷ A systematic review by Singer et al. identified a well-organized transition, a treatment summary, a survivorship care plan (SCP), the education of generalist providers, and guidelines as components of successful followup.²⁸

We identified 41 resources available to both survivors and providers (Appendix C). Of these, 25 resources are survivor-specific, 10 are provider/researcher-specific, and 6 are meant for use by both. We also identified eight U.S. guidelines (7 physician-specific, and 1 survivor-specific), and 6 international guidelines (5 physician/researcher-specific, and 1 survivor-specific). Based on feedback from the Stakeholders we identified resources that were free to both survivors and providers (Appendix C). According to our Stakeholders, resources are most helpful if they are easy to access, user-friendly, known to survivors and providers, from trustworthy sources, and valued by survivors and their families (e.g., asking for updates to treatment summary).

Development of Initial Program Theory

We developed an initial program theory based on input from the Stakeholders and a review of the literature. The final products from this initial process are a variable list and initial program theory, which are shown in Appendix D and Figure 2, respectively. Below, we describe the iterative process that produced these final products. In our discussions with Stakeholders we used the term ‘patient’ but use the term ‘survivor’ throughout the report.

Discussion with Stakeholders

In our first round of discussions the Stakeholders provided their input on (1) what models of care and resources are available to support survivorship care for childhood cancer survivors and (2) how effectiveness is defined for these models and resources. Based on this initial round of discussions, we also determined that this realist review would focus on adult survivors of childhood cancer only, as the models of care and resources for survivors while still children or adolescents are substantially different.

The Stakeholders identified a number of factors that influence the delivery of survivorship care to childhood cancer survivors. The fractured U.S. health care system and availability of financial and other resources were identified as key variables at the system, provider, and survivor level. In addition, coordination of care – among primary care and specialty providers, and between providers and survivors – influences the care that is delivered/received and whether and when transition to primary care occurs. The Stakeholders also noted the importance of survivor-specific characteristics, such as developmental age and time since diagnosis, complexity of the diagnosis and treatment(s), current effects of treatment, and risk for late effects.

In terms of effectiveness, the Stakeholders identified mortality, morbidity, quality of life, and costs as the key final outcomes, summarized as “Survivors live longer and feel better through high-value care.” They also noted a number of intermediate outcomes, including loss to followup; surveillance for late effects and concordance with guidelines; communication, transfer of information, and care coordination; returning survivors to an informed PCP/helping survivors identify knowledgeable providers; ensuring survivors know what they need; and connecting survivors with resources and services. Some additional factors related to resource effectiveness include that they be easy to access, user-friendly, known, and trusted. In combination, the intended effects of the models of care and resources were summarized as “Survivors feel confident about sharing their history, know their risks, recognize symptoms and problems,

understand the care they need, are aware of the resources available to help them, and can access relevant care and services.”

Literature Review

We searched the literature for existing mid-range theories that could inform our understanding of the relationships among the key factors identified by the Stakeholders. We also reviewed editorials, commentaries, qualitative, and mixed methods studies to further elucidate our understanding of the key factors, as well as the purported mechanisms and outcomes, for childhood cancer survivorship models of care and resources.

The Andersen Behavioral Model of Health Services Use²⁹ best fit the identified factors from the initial discussions with Stakeholders. In particular, this version of the Behavioral Model of Health Services Use describes how environmental factors (health care system, external environment) relate to population characteristics (predisposing characteristics, enabling resources, need) that influence health behavior (personal health practices, use of health services) that lead to outcomes (perceived health status, evaluated health status, consumer satisfaction).

With minor modifications, we were able to fit the key factors identified by the Stakeholders into the Andersen model categories. Modifications included referring to “individual characteristics” rather than “population characteristics,” renaming “enabling resources” as “facilitators/barriers,” separating survivor and provider health behaviors, and adding “cost” as an outcome.

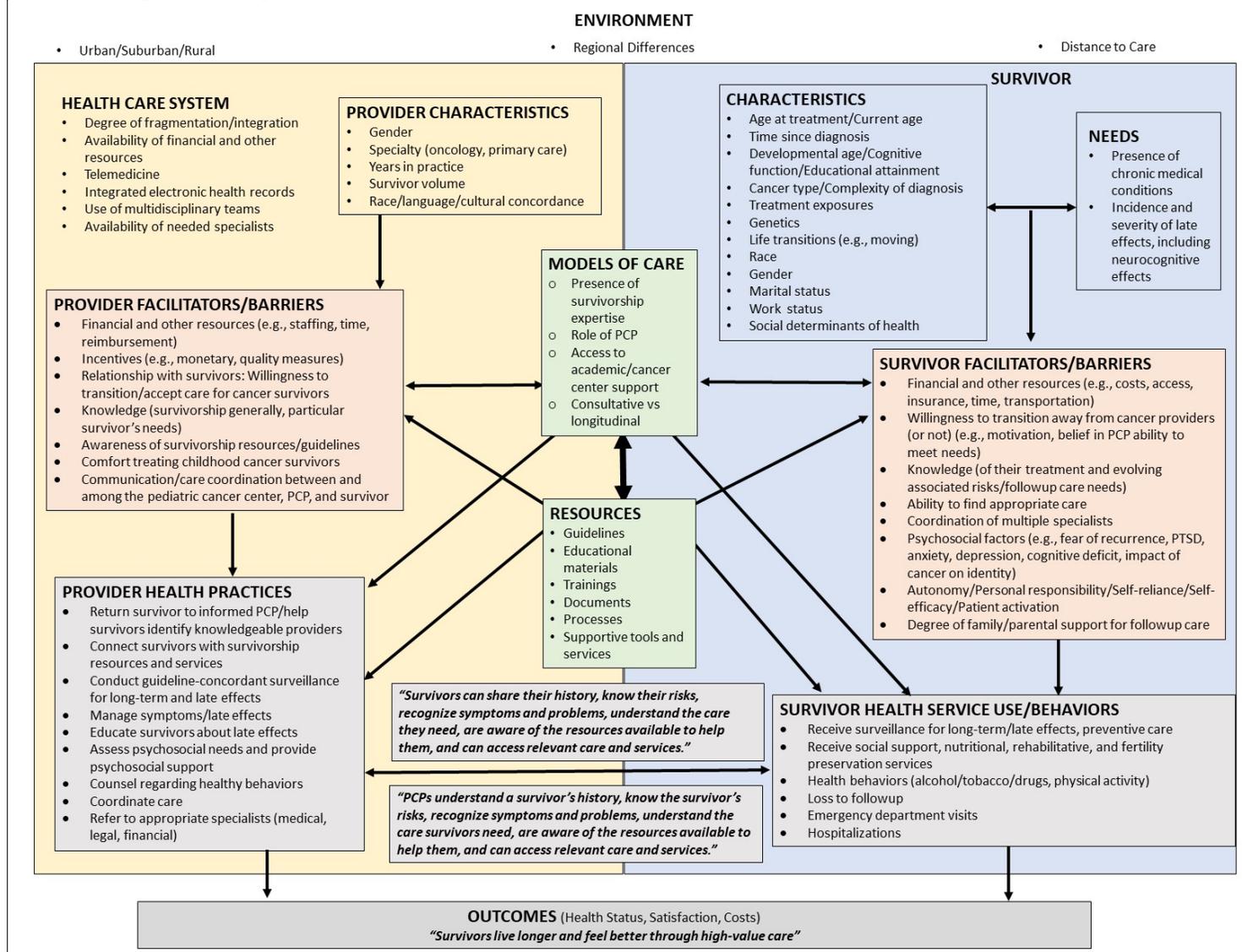
We then organized our empiric literature review according to the categories from the modified Andersen model (e.g., environment, individual characteristics, health behaviors, and outcomes). The goal was to identify variables in each of these categories that describe and affect the contexts, mechanisms, and outcomes of childhood cancer survivorship models of care and resources.

Developing Initial Program Theory

As shown in Appendices D and E (initial program theory iterations) and in Figure 2 (refined program theory), we have separated the health system/provider factors (shown in yellow) from the patient factors (shown in blue), with the overall environment (shown in white) as the background. The models of care and resources sit at the intersection of health system/provider factors and patient factors and are shown in green. We describe the health care system attributes and provider factors in yellow and the patient characteristics and needs in blue. The modifiable facilitators barriers on both the health system/provider side and survivor side are shown in peach. Intermediate outcome and process measures (provider health practices; survivor health service use/behaviors) are displayed in light gray, with the final outcomes displayed in dark gray. Each of these aspects of the program theory are described in more detail below.

On the health system/provider side, health care system factors (e.g., degree of fragmentation and integration, availability of financial and other resources, availability of needed specialists) set the stage for provider factors, including characteristics, facilitators/barriers, and practices. Key provider characteristics include specialty (e.g., oncology, primary care) and years in practice. The provider facilitators and barriers of survivorship care include financial and other resources (e.g., staffing, time, reimbursement); providers’ relationships with survivors; willingness to transition/accept care for cancer survivors; knowledge about survivorship, generally, and particular survivor’s needs; awareness of survivorship resources/guidelines; comfort with treating childhood cancer survivors; and communication/care coordination between

Figure 2. Refined Program Theory.



PCP = primary care provider; PTSD = post-traumatic stress disorder.

and among providers and survivors. Provider health practices include returning survivors to an informed PCP/helping survivors identify knowledgeable providers, connecting survivors with resources and services, conducting guideline-concordant surveillance for long-term and late effects, managing symptoms/late effects, educating survivors about late effects, assessing psychosocial needs and providing psychosocial support, counseling survivors on healthy behaviors, coordinating care, and referring to appropriate specialists (medical, legal, financial).

On the survivor side, key characteristics identified include age at treatment and developmental age, time since and complexity of diagnosis and treatment, race and gender, life transitions (e.g., moving), and genetics. Key needs are the presence of chronic medical conditions and the risk for/severity of late effects. Survivor facilitators/barriers of survivorship care include financial and other resources (e.g., insurance, time, transportation), willingness to transition away from cancer providers, knowledge about their treatment and followup care needs, psychosocial factors (e.g., fear of recurrence, anxiety, depression), autonomy/personal responsibility/self-reliance, and degree of family/parental/partner support for followup care. Relevant survivor health services use includes receiving surveillance for long-term/late effects and preventive care; receiving social support, nutritional, rehabilitative, and fertility preservation services; emergency department visits and hospitalizations; and whether the survivor is lost to followup. Health behaviors include use of alcohol/tobacco/drugs and physical activity.

The provider health practices and survivor health services/use behaviors represent the intermediate outcomes as described by the Stakeholders “Survivors feel confident about sharing their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services.” These outcomes lead to the final outcomes of health status, satisfaction, and costs, or as described by the Stakeholders, “Survivors live longer and feel better through high-value care.”

This initial program theory was shared with the Stakeholders in a second round of discussions. The Stakeholders broadly supported the theory and felt that it accurately and comprehensively described the influence of models of care and resources for childhood cancer survivors. They suggested minor additions to the list of variables. For example, on the health-system/provider side, they noted the impact of crisis events such as the COVID-19 pandemic, and how these can influence care delivery in important ways. They suggested that concordance of race, language, and culture between providers and survivors could be important and recommended that the general category of survivor social determinants of health be added. Regarding financial aspects, they noted that incentives (both financial and related to quality programs) could play a role on the provider side and that financial costs on the survivor side include reimbursement and ability to pay their co-pays. For survivors, coordination of multiple specialists was identified as a particular challenge. Other comments primarily related to the refinement of how variables were listed.

Refinement of Initial Program Theory

To refine the initial program theory, we sought empiric evidence and evaluated whether the evidence supported, refuted, or suggested revisions to our initial program theory. Here, we describe how we updated the variable list (Appendix D) and revised the initial program theory (Figure 2) based on the empiric literature.

There was at least some evidence addressing almost all of the variables in our initial list. The variables most commonly found in the literature we reviewed included oncology versus primary care; cancer type and complexity of diagnosis; survivor age, race, and gender; survivor and

provider financial and other resources; survivor and provider knowledge; provider comfort with treating childhood cancer survivors; communication and coordination between and among providers and survivors; delivery/receipt of prevention and surveillance of long-term and late effects; and quality of life/health status and satisfaction. Variables that were seen less frequently include crisis events, genetics, emergency department visits, hospitalizations, and costs.

The literature also identified some factors that we added to our list of variables. Under Environment, we added regional differences. Under Provider Characteristics, we added gender and survivor volume. Under Survivor Characteristics, we added cancer type, current age, marital status, and work status; we also clarified that time since diagnosis represents both longitudinal (5 years vs. 10 years from diagnosis) and calendar (e.g., diagnosed in 1985 vs. 1999) time. Under Survivor Facilitators/Barriers, we added awareness and availability of (culturally appropriate) resources.

The most important substantive change made to the initial program theory was the addition of an intermediate outcome focused on PCPs. Specifically, our initial steps had identified as the intermediate outcome that “Survivors feel confident about sharing their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services.” The evidence in the literature demonstrated that there is also an important provider component as an intermediate outcome, which is “PCPs understand a survivor’s history, know the survivor’s risks, recognize symptoms and problems, understand the care survivors need, are aware of the resources available to help them, and can access relevant care and services.”

In the final round of Stakeholder discussions, we reviewed these findings with the Stakeholders, who reported that our results were consistent with their understanding based on their experiences with and expertise in childhood cancer survivorship care.

Realist Review Key Questions 1 and 2

KQ1. For whom and under what circumstances could different survivorship care models for adult survivors of childhood cancer (cancer diagnosed prior to age 21 years old) that include primary care be effective?

- a. What are the key mechanisms by which these models could be effective?
- b. What are important contexts that determine whether different mechanisms could be effective?

KQ2. For whom and under what circumstances could different survivorship care resources for adult survivors of childhood cancer be effective in achieving their intended outcomes?

- a. For survivors and their families
 - i. What are the key mechanisms by which these resources could lead to their intended outcome?
 - ii. What are important contexts that determine whether different mechanisms could lead to outcomes?

- b. For care providers
 - i. What are the key mechanisms by which these resources could lead to their intended outcome?
 - ii. What are important contexts that determine whether different mechanisms could lead to outcomes?

Development of Context-Mechanism-Outcome Hypotheses

To address the realist review KQs related to the contexts and mechanisms associated with effective models of care that include primary care (KQ1) and of resources (KQ2) that achieve their intended outcomes, we abstracted information supporting or refuting our program theory from the empiric literature and developed seven CMO hypotheses. Because the CMOs related to the models of care and resources were closely linked, we addressed these KQs jointly. We also noted that the realist review KQ regarding models of care was not *whether* models of care that include primary care are effective for adult survivors of childhood cancer but, rather, in what contexts and via what mechanisms they *could* be effective.

These CMO hypotheses were primarily developed based on evidence from literature describing childhood cancer survivors (both during childhood/adolescence/young adulthood and adulthood), though we also drew from evidence regarding adult survivors of adult cancers where relevant and informative. For each CMO, we provide example evidence from the literature that supports, and in some cases, refutes, the hypothesis. As with all realist reviews, the evidence is provided for illustrative purposes and is not intended to be a comprehensive summary.

CMOs Focused on Survivor Intermediate Outcome

Four CMO hypotheses focus on the survivor intermediate outcome of “Survivors feel confident about sharing their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services,” as shown in Table 2.

Table 2. CMO hypotheses focusing on the Survivor intermediate outcome.

In the CONTEXT of	this MECHANISM ... produces	OUTCOME (intermediate)	OUTCOME (final)
A) the availability of survivorship care plans, guidelines, and other resources	A1) improved survivor knowledge A2) information available to share with PCP to inform delivery of survivorship-related care	Survivors can share their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services.	Survivors live longer and feel better through high-value care.
B) healthier survivors (perceived or actual)	B) less perceived/actual need for survivorship-related care		
C) survivors engaged in health care system	C) improved knowledge		
D) survivor confidence in PCPs	D) willingness to transition care		

CMO = context-mechanism-outcome; PCP = primary care provider.

Context A: SCPs, guidelines, and other resources

In the literature, we found that the availability of SCPs, guidelines, and other resources led to the survivor-focused intermediate outcome via two mechanisms: (1) survivors have improved knowledge regarding their care needs and (2) survivors have information to share with their PCPs to inform delivery of survivorship-related care.

We also identified four sub-themes related to the sharing of information through these resources: (1) improved knowledge can be both beneficial and detrimental, (2) resources have bigger impacts on survivors with lower knowledge at baseline, (3) issues regarding resource “dose,” and (4) gender differences in engagement with and impact of resources.

Mechanism A1: Improved knowledge for survivors

Regarding the first mechanism, in a survey of 1,395 adolescent/young adult (AYA) survivors, Shay et al. found that SCPs and/or followup care instructions were significantly associated with lower odds of survivors reporting unmet needs around information about topics such as late effects of treatment, fertility, and recurrence concerns.³⁰ As another example, childhood cancer survivors 2-years post-treatment who were given a passport card describing diagnosis, treatment, risks, and recommended followup were more likely to demonstrate improved knowledge versus survivors without a passport.³¹ A third study, conducted by Yan et al., found that possession of an SCP among high-risk survivors was associated with increased adherence to breast, skin, and cardiac surveillance.³²

Mechanism A2: Transfer of information to PCPs

Regarding the second mechanism, there was also evidence that survivors valued the information from SCPs and other resources to share with their other providers. For example, in a study of 111 adult survivors of pediatric and young adult cancer, 95 percent reported understanding everything on the treatment summary, 83 percent found it extremely or moderately valuable, and 95 percent found the SCP helpful in understanding the plan for their care.³³ In addition, of 30 respondents who had visited an outside provider since getting the SCP, one-third gave the provider a copy of the form and 44 percent gave a copy to someone in their personal circle. However, intentions to share the SCP do not always translate into action. In a cohort study of 20 families of a child with acute lymphoblastic leukemia, 95.75 percent of parents reported intending to share the SCP with someone, but only 60.9 percent had done so at the third followup, and only 35 percent of those with a PCP had shared it with them by Time 3.³⁴

In a prospective one-arm study assessing SCP and information provision in 62 survivors at increased risk for late effects, nearly half of those who had seen a physician since SCP receipt had shared it; most who hadn't seen a physician planned to share the SCP when they did.³⁵ Further, the investigators found that PCPs seemed too busy to be involved in the study or to utilize the resources offered to them directly, but they ordered tests when the survivor presented them with information on risks. In a study of 5,661 adult survivors of childhood cancer, Steele et al. found that discussing cancer-related risks with a doctor is the strongest predictor of getting screened for late effects and that the physician's access to the survivor's cancer treatment summary significantly predicted screening for relevant health risks.³⁶ Based on these findings, the authors discuss the importance of communication between survivors and providers regarding receipt of appropriate care and the role of resources, such as treatment summaries, in promoting this communication.

Subtheme 1: Benefits and harms of information. An important theme in the literature about resources and the information they provide is that they can be both beneficial and detrimental. For example, the Spain et al. study of adult survivors of pediatric and young adult cancer found that 14 percent reported being concerned by the SCP as a whole, and 28 percent were concerned by potential late effects.³³

A randomized controlled trial (RCT) of SCPs in gynecologic cancer survivors (not childhood cancer survivors) investigated this issue in greater depth. In this Dutch study of an electronic

medical record-generated SCP for survivors of ovarian and endometrial cancer, endometrial cancer survivors in the SCP arm reported greater concern about their illness, more emotional effects, and more symptoms.³⁷ Similarly, the ovarian cancer survivors in the SCP arm reported less trust that the treatment would cure their disease.³⁸ The authors noted that these negative outcomes are not necessarily bad. For example, the endometrial cancer survivors had more cancer-related contact with their PCPs, which the authors speculated relates to the survivors' greater awareness of cancer-related symptoms and the possibility that having the SCPs empowered them to seek out the necessary support.³⁷ They also suggest that health care providers may be reluctant to share information about potential late effects to avoid such negative consequences, but that avoidance of the issue may limit the survivor's awareness and empowerment. For the ovarian cancer survivors, the authors note that the decreased belief in the potential for cure gleaned from the SCP may be more realistic, but that it is an issue providers may be reluctant to address.³⁸

In an analysis of 7,917 cancer survivors (unspecified if childhood or adult cancer survivors) who participated in the Cancer Survivorship Module of the 2010 Behavioral Risk Factor Surveillance System, Jabson et al. found a similar association. Specifically, they found that survivors who had received a treatment summary, followup care instructions, or both were more likely to report pain.³⁹ The authors note that the documents might heighten cancer survivors' awareness and report of cancer-related pain; alternatively, it could be that patients who undergo more extensive treatment are more likely to both experience pain and receive a treatment summary and followup care instructions. A different analysis by Jabson, this one in 3,541 cancer survivors who participated in a 2010 LIVESTRONG online survey, had different findings.⁴⁰ In this population, they found that survivors who received followup care instructions reported 25 percent fewer late effects. In this paper, they hypothesized that cancer survivors who receive followup care instructions become more aware of the symptoms of late effects sooner, leading them to obtain care earlier and resulting in them reporting fewer problems.

The authors of the Dutch RCT conducted a followup analysis that examined how different preferences for information affected the impact of SCPs.⁴¹ They compared "monitors," who desire information about their disease to "blunters," who avoid information. SCPs were more beneficial to monitors across the board, but particularly those who did not have easy access to other information sources, such as the internet. For blunters, those in the SCP arm reported a greater impact of the disease on life and more concerns about the illness compared with blunters in the control arm.

All of these studies raise the question whether delivery of information should be tailored to the preference of the survivor.

Subtheme 2: Resources have greater impact for survivors with lower baseline knowledge. Several studies have found that resources improved knowledge more in survivors who knew less. For example, in one study, new patients were more likely to report learning new information from the SCP compared with return patients.³³ In a different study, which tested a survivorship clinic visit intervention in 369 adult survivors of childhood cancer, survivors with the lowest knowledge of therapy and therapy-related health risk at baseline had the greatest gains.⁴² Papers with similar findings in adult survivors of adult cancers have posited that the failure of resources to demonstrate impacts may be due in part to high levels of knowledge and/or few or no needs in the populations being studied.^{43, 44}

These findings raise the question of whether resources should be targeted to survivors who have information deficits or needs and, as described above, for whom the resource will be beneficial rather than detrimental.

Subtheme 3: Resource “dose.” An important consideration in implementing resources is that they deliver a sufficient “dose” to be effective. Several studies noted that failure to find an effect of the studied resource may have been related to an insufficient dose. For example, Hudson et al. found that a brief, broad-based risk counseling intervention did not achieve a substantial long-term change in knowledge, health perceptions, or health practices.⁴⁵ Similarly, Steele et al. found that their “relatively weak intervention dose” of a targeted (not tailored) page of information in a newsletter did not lead to increased medical followup in at-risk pediatric cancer survivors. They also noted that tailored information is more consistently effective, though effects are small.³⁶ As an example from studies of adult survivors of adult cancers, Turner et al. said that timing or insufficient “dose” may have led to the null findings in their trial of a Head and Neck Cancer Survivor Self-Management Care Plan.⁴⁶

Based on this evidence, one may consider implementing more intensive interventions. However, as discussed below regarding providers, practical considerations limit how much time and effort a resource can involve.

Subtheme 4: Gender differences. One final subtheme identified in the literature was differences in gender regarding engagement with and impact of resources. For example, Hudson et al. found that females had a statistically significantly greater improvement in knowledge following a multi-component risk-counseling intervention versus males, though the difference was small.⁴⁵ Similarly, in the Steele et al. study comparing two approaches for sharing information via a newsletter, more women (72 percent) than men (59 percent) reported reading the newsletter.³⁶ In the Oeffinger et al. single-arm study, more women than men visited the study website that provided survivorship resources.³⁵ These results raise questions regarding whether resources should be tailored to different groups (e.g., males vs. females) based on how they engage and use them.

Context B: Survivors who perceive themselves to be, or are actually, healthier

A different context in which models of care that include primary care could be effective for childhood cancer survivors is when survivors perceive themselves to be – or are actually – healthier. This CMO hypothesis is consistent with the NAM report’s recommendation for risk-adjusted followup care.⁸

In one analysis of 6,176 survivors from the Childhood Cancer Survivor Study, survivors who reported no morbidity at baseline were less likely to report receiving care at followup, whereas survivors who reported any chronic health condition at baseline were more likely to report care at followup.⁴⁷ Another analysis of the Childhood Cancer Survivor Study found that survivors who received more intensive therapy; those with a severe, life-threatening, or disabling chronic condition; and those who reported worse cancer-related pain were more likely to have received care.⁷ Klosky et al. found that one factor associated with non-attendance at the St. Jude followup clinic was having had no additional cancer event.⁴⁸

In a survey of 160 Swiss AYA survivors, non-attenders of followup were more likely to rate models of care involving a GP or via telephone/questionnaire higher than attenders.⁴⁹ Non-attenders were also less likely to report late effects than attenders of followup. In another study, low perceived need for care was a key factor highlighted for not engaging in care.⁵⁰ Guilcher et al. conducted a survey of 16 pediatric hematology/oncology programs regarding long-term

followup care programs for childhood cancer survivors in Canada. They found that the key factors associated with followup in a formal adult late effects program were existing late effects, higher risk for adult onset late effects, and time from completion of therapy;⁵¹ one would expect the converse factors to be associated with followup in primary care (or, perhaps, no followup).

This evidence raises two issues regarding the risk-based followup care recommended by the NAM. First, attendance at clinic is not equivalent to need for care. Some low-risk survivors may be unwilling to transfer care if, for example, they do not have confidence in their PCP to provide care (as described below). Second, non-attendance at clinic is not equivalent to no need for care. Some of the survivors classified as non-attenders may not have been fully aware of their risk for late effects. This theory is supported by an RCT that investigated a tailored counseling intervention in 472 Childhood Cancer Survivor Study participants at-risk for cardiomyopathy.⁵² While more survivors (52.2 percent) in the intervention arm completed cardiomyopathy screening compared with controls (22.3 percent), one of the reasons cited for not getting screened was not perceiving it as important. Notably, screening detected cardiomyopathy or other abnormalities consistent with evolving cardiac dysfunction in over 20 percent of those screened.

Context C: Survivors who are engaged in the health care system

Based on the literature, being engaged in the health care system (e.g., physician visits) provides another context in which survivors can gain the knowledge they need and receive appropriate care, though certainly not universally. For example, among 106 young adult survivors of childhood cancer, 63.2 percent reported that, during their last clinic visit, a provider discussed any symptoms related to their cancer, with less than half reporting that they promoted adherence to care recommendations, interest in reproduction, social support, and mental health issues.⁵³ A Swedish study of 213 young adult survivors of childhood acute lymphoblastic leukemia found that respondents who had no regular contact with health care services were more likely to report that they had not received knowledge, treatment strategies, or guidance for coping with physical changes.⁵⁴ In addition, satisfaction with level of contact was associated with reporting receipt of knowledge and treatment strategies regarding physical changes – those who were more satisfied were more likely to report receipt. Nevertheless, even among the respondents who were satisfied with the level of contact, only a minority reported receiving sufficient knowledge and treatment strategies about physical changes. Regarding receipt of care, Yan et al. found that having a cancer-related check-up in the past 2 years was associated with increased surveillance for breast, skin, and cardiac late effects; visiting a doctor more than five times in the past 2 years was associated with increased surveillance for skin and cardiac late effects.³² The sociodemographic factors consistently associated with *not* being engaged in care include male sex, lack of insurance, lower income, race (non-White or other), and less education.^{7, 47, 48, 53}

These findings provide insight regarding the pathways to appropriate receipt of care; it seems that survivors who are engaged with the health care system learn more about their care needs and are more likely to receive appropriate care.

Context D: Survivors who have greater confidence in their PCPs

A final context describes how survivors who are confident in their PCPs might be more willing to transition their care. In the Stakeholder discussions, one of the Stakeholders noted that

it is important that the survivor’s “emotional/physical escorts” (e.g., their parents or partners) also be confident in primary care for the transition to occur.

The evidence regarding this CMO hypothesis actually supports the converse: survivors are not confident in PCPs and prefer models of care that include cancer specialists. For example, in an Australian survey of 633 parents of childhood cancer survivors, AYA survivors, and older survivors of childhood cancer, hospital-based survivorship care—involving an oncologist or other clinic doctor, cancer survivorship nurse, or team—was the first choice of 97 percent of parents, 88 percent of AYAs, and 86 percent of older survivors.⁵⁵ They had lower confidence in PCPs. Similar findings were seen in a survey of 160 Swiss AYA survivors, who rated medical oncologist involvement most highly for survivorship care.⁴⁹ The authors noted that, even though the respondents were surveyed at least 5 years after diagnosis, their biggest concerns were cancer relapse and occurrence of late effects. They speculate that survivors may perceive medical oncologists as best suited to deal with these issues. An alternate perspective is provided from a survey in the Netherlands of 133 adult survivors of childhood cancer.⁵⁶ In this study, 88 percent of survivors were satisfied with the care given by local family doctors, and only 14 percent thought their local family doctor’s knowledge of their medical history was inadequate.

The limited evidence regarding this CMO hypothesis suggests that work is required to increase the confidence of cancer survivors in PCPs to facilitate models of care that include primary care.

CMOs Focused on Provider Intermediate Outcome

Three of our CMO hypotheses focus on the provider intermediate outcome of “PCPs understand a survivor’s history, know the survivor’s risks, recognize symptoms and problems, understand the care survivors need, are aware of the resources available to help them, and can access relevant care and services,” as shown in Table 3. Several Stakeholders commented that all three CMO hypotheses relate to PCPs obtaining the knowledge needed to appropriately care for childhood cancer survivors – either through SCPs, guidelines, and resources; shared-care with an oncologist; or greater experience caring for childhood cancer survivors.

Table 3. CMO hypotheses focusing on the provider intermediate outcome.

In the CONTEXT of	this MECHANISM ... produces	OUTCOME (intermediate)	OUTCOME (final)
A) the availability of survivorship care plans, guidelines, and other resources	A) information available to guide the PCP in delivering survivorship-related care	PCPs understand a survivor’s history, know the survivor’s risks, recognize symptoms and problems, understand the care survivors need, are aware of the resources available to help them, and can access relevant care and services.	Survivors live longer and feel better through high-value care.
B) shared-care with oncologist	B) support from the oncologist to aid the PCP in delivering survivorship-related care		
C) more experience caring for childhood cancer survivors	C) greater comfort caring for childhood cancer survivors		

CMO = context-mechanism-outcome; PCP = primary care provider.

Context A: SCPs, guidelines, and other resources

For providers, we found that the availability of SCPs, guidelines, and other resources work through the mechanism of having information available to guide the PCP in delivering survivorship-related care.

Multiple studies have documented the value that PCPs place on resources to support their delivery of survivorship care. In one survey, internists (n=1,110) and family practitioners (n=1,024) rated highly long-term followup guidelines and survivor-specific standardized letters from specialists with followup recommendations for supporting their delivery of survivorship care.^{26, 27} Similarly, another survey of 351 general internal medicine and family practice providers found that more than 90 percent thought it would be useful to receive a treatment summary or SCP, and 86 percent agreed they would follow guidelines.⁵⁷ In a cross-sectional survey of 27 medical oncologists, 13 pediatric oncologists, 122 GPs, and 21 pediatricians in Switzerland, all groups reported a need for standardized protocols (85 to 91 percent) and specialized training (55 to 73 percent).⁵⁸ These resources are also endorsed as a key enabling factor for shared care. Among 233 GPs in the Netherlands who had taken a postgraduate course on late effects of cancer treatment, two of the main requirements for participation in shared care were availability of guidelines (64 percent) and sufficient information about the survivor's medical history (37 percent).⁵⁹

There is also some evidence that PCPs use these resources and that the resources promote quality survivorship care. Among PCPs who recalled receiving an SCP as part of a research study, 75 percent reported often or always reviewing the plan, and 42 percent reported discussing the SCP with the survivor; further, 48 percent found the treatment summary very helpful, and 56 percent found the followup surveillance recommendations helpful.⁶⁰ Yan et al. found that PCP possession of an SCP was associated with increased adherence to breast and colorectal screening among survivors at average risk, though less than half of high-risk survivors received recommended surveillance.³²

Despite the potential value of these resources, there is also evidence that they currently have limited reach and effectiveness. In a survey of 351 US general internal medicine and family practice providers, respondents endorsed the value of SCPs and guidelines, but approximately 85 percent reported never receiving a cancer treatment summary or SCP, and 93 percent reported never using the Children's Oncology Group (COG) late effects guidelines.⁵⁷ Further, only 40 percent of providers were aware of the guidelines, and less than 40 percent felt their training was adequate to recognize late effects. However, Yan et al. reported substantial improvements in adherence to the COG guidelines between 2003 and 2016 and speculated that physician awareness of COG guidelines may be growing.³² In a study of SCPs published in 2017, a survey of 134 PCPs found that only 31 percent of the PCPs felt very comfortable providing the surveillance recommended on the SCP, 19 percent felt very comfortable using the SCP to provide recommended surveillance for neurocognitive late effects, and 3 percent felt very comfortable providing all aspects of survivorship care.⁶⁰ In discussing these results, the authors noted the limitations of passive SCP distribution and the need to investigate ways to deliver information to PCPs to better address their knowledge needs.

This evidence suggests that there is potential value for SCPs, guidelines, and other resources in promoting effective childhood cancer survivorship care, but that their potential has yet to be fully realized. As relates to this point, we identified three sub-themes: 1) lack of awareness of the resources; 2) role of the electronic health record (EHR) in managing information; and 3) tension between resources delivering a sufficient "dose" and practical considerations regarding what PCPs can take the time to do.

Subtheme 1: Lack of awareness of resources. An interesting finding from the SCP study of Iyer et al. was that, even though all PCPs were sent an SCP as part of the study, only 51 percent recalled receiving it.⁶⁰ More information on this point is found in the literature regarding adult

survivors of adult cancer. Donahue et al. obtained the perspectives of 81 PCPs seeing cancer survivors enrolled in a survivorship clinical trial.⁶¹ PCPs reported that the main barriers to SCP use were not knowing a plan existed (83 percent), not knowing how to find the plan (75 percent), not being able to locate the plan in the patient chart (71 percent), and not knowing to look for the plan (72 percent). Notably, these barriers were reported even though the SCPs are standardly housed in the EHR problem list within their institution. Potential facilitators identified by the PCPs included awareness of plan existence (85 percent), a standardized location within medical records (89 percent), and consistent provision of care plans for all patients (81 percent). The PCPs noted that an SCP designed specifically for them, rather than one designed to serve both survivors and PCPs, may be more helpful. In discussing the findings, the investigators speculated that their institution may have yet to reach a “critical mass” of SCP provision that would facilitate PCPs’ awareness of and ability to use them. They suggest the need for “primary care-centered design of SCP format and content, location in the EHR, and the ability to ‘push’ relevant or needed survivorship information to primary care at the right time.” Another study that included 21 physicians or dentists of adult survivors of adult cancer found that only 34 percent recalled receiving the SCP or could locate it.⁶² The authors of this study compared the SCP to “a needle in a haystack” for health care professionals across institutions who use different EHRs or paper charts.

Knowing that an SCP exists is a critical first step in being able to use it in practice, and EHRs can both help and hinder that process. Below, we discuss the role of EHRs in creating SCPs.

Subtheme 2: Role of EHRs. A number of studies of adult survivors of adult cancers commented on or highlighted the role of the EHR in creating SCPs. The SCPs used in the Netherlands RCT in gynecologic cancer were auto-populated from the EHR,^{37, 38, 63} as were the SCPs used in a study of hematopoietic stem cell transplantation survivors.⁶⁴ Morken et al. describe the advantages of using EHRs to create and provide SCPs, including reduced time and resources to compile the information, producing a document that is electronically searchable, and facilitating updates to the plan when needed.⁶⁴ However, they also noted that, for EHRs to support the effective use of SCPs, discrete data capture is required, organization policies and technologies must be designed to support clinician needs, and survivorship-related tasks need to be clearly assigned. Perhaps because of these barriers, in surveys of cancer programs and cancer care providers, Birken et al. found that EHR systems used to create SCPs were lacking and/or underused.^{65, 66}

The potential role of EHRs to promote the creation, delivery, and data management of SCPs requires further exploration.

Subtheme 3: Resource “dose.” Similar to the subtheme described above regarding survivors, there is also a tension between resources delivering a sufficient “dose” to providers and practical considerations regarding what PCPs can take the time to do. For example, as part of a survivorship study, Oeffinger et al. developed a virtual information center for childhood cancer survivors and their providers, but none of the PCPs who agreed to participate in the study visited the website.³⁵ In a different study, Costello et al. aimed to implement a telemedicine transition visit with a PCP and childhood cancer survivors joined by a pediatric survivorship clinic team member.⁶⁷ However, only 24 of 46 eligible dyads agreed to participate, due in part to PCPs’ hesitation with using the study-provided telemedicine equipment. From the adult survivorship literature, there is some evidence that providers prefer more targeted information, such as SCPs that are shorter and directed to PCPs’ needs, rather than information directed to both PCPs and

survivors.⁶¹ One Stakeholder raised the issue of whether providers might be more willing to undertake these activities if reimbursement were sufficient.

Similar to what was found with survivors, there is a delicate balance between providing a sufficient “dose” of information to be effective and being realistic regarding the time and effort providers can invest.

Context B: Shared-care with oncologist

Similar to the reports from survivors above regarding preferences for models of care that include cancer specialists, providers highly endorse shared-care models. Among 1,110 internists and 1,024 family practitioners, 84 percent and 85 percent, respectively, prefer working in collaboration with a cancer center-based physician or long-term followup clinic.^{26, 27} Among the 134 PCPs interviewed in another study, 46 percent selected specialty survivorship clinic as the most preferred model of care, followed by 26 percent preferring shared care.⁶⁰ In a questionnaire completed by 233 Dutch GPs who had taken a postgraduate course on late effects of cancer treatment, 97 percent were willing to participate in a shared-care model for followup.⁵⁹ Among these respondents, in addition to endorsing the importance of guidelines and relevant information about the survivor's medical history (described above), they also noted the importance of short communication lines to support shared care. In a separate study, these authors conducted a survey of 133 adult survivors of childhood cancer and 115 family doctors of adult survivors of childhood cancer regarding shared care and found that 82 percent of participating local family doctors were satisfied with the shared care collaboration and thought the information they had received from the long-term followup clinic was adequate.⁵⁶ In this study, all family doctors were provided information on the survivor's history, health risks, and required tests.⁵⁶ Other studies have also noted the importance of regular communication and close collaboration between pediatric oncology and PCPs, as well as documents such as SCPs, so that PCPs are aware of and can address the unique health risks of childhood cancer survivors.^{49, 52} However, there is some evidence that current levels of communication are inadequate. In a Swiss survey of providers, 94 percent of GPs reported wanting more support from oncologists.⁵⁸

Two studies from the literature related to adult survivors of adult cancer provide additional insights. In a description of a Primary Care for Cancer Survivor Clinic, Choi et al. report how the high quality of communication between the clinic and oncology providers is valued.⁶⁸ Another study looked at the degree to which providers shared patients in a cohort of 8,661 cancer survivors and found that a greater degree of patient sharing among providers was associated with higher quality care on some measures and lower costs.⁶⁹

When implemented effectively, shared-care models provide close connection and quality communication so that PCPs have the information they need from cancer specialists to deliver appropriate survivorship care.

Context C: More experience caring for childhood cancer survivors

There is limited evidence suggesting that the more experience PCPs have caring for cancer survivors, the more comfortable they feel doing so and the better they adhere to guidelines. In a survey of 1,110 internists, those who saw at least one childhood cancer survivor in the last 5 years reported being more comfortable seeing childhood cancer survivors and were more likely to promote appropriate breast cancer surveillance.²⁷ Findings in a survey of 1,024 family practitioners were similar.²⁶ Family physicians who had seen at least one cancer survivor in the

past 5 years reported more comfort in doing so and were more likely to correctly identify appropriate surveillance strategies.

However, most PCPs treat few if any childhood cancer survivors. Based on data from two surveys, only 51 percent of internists and 58 percent of family practitioners report having cared for one or more childhood cancer survivors in the past 5 years.^{26,27} In another survey of PCPs, 40 percent reported they had never cared for one.⁵⁷ Among the 134 PCPs in the Iyer et al. study, the average number of childhood cancer survivors cared for in the past 5 years was one, with 84 percent reporting having cared for only one.⁶⁰ They also had little exposure to late effects, with only 34 percent having seen at least 5 late effects and 45 percent reporting seeing at least one late effect of grade 3 or higher. In Switzerland, Michel et al. discuss the possibility of having one specialized PCP for a certain catchment area, although the possibility of translating this idea to the U.S. context is unclear.⁵⁸

These themes were echoed in a study of 86 PCPs regarding care for adult survivors of hematologic malignancies and hematopoietic cell transplantation.⁷⁰ PCPs who had cared for more survivors felt more confident and perceived fewer barriers to doing so. They also were more likely to report that they discussed screening and late effects with patients.

While there is some evidence that having a greater concentration of childhood cancer survivors in a PCP's practice could promote greater provider comfort and lead to more appropriate care, opportunities to operationalize this approach in the U.S. may be limited.

Discussion

Summary of Findings

This realist review addresses how models of care that include primary care, and resources for adult survivors of childhood cancer and their families and providers, can be effective. Undergirding this review were CQs that define effectiveness for the models and resources; describe models of care, highlighting those that include primary care; and identify resources directed to childhood cancer survivors and their families and to care providers.

Effectiveness for both resources and models is defined by survivors living longer and feeling better through high-value care. Intermediate measures of effectiveness evaluate the extent to which (1) Survivors feel confident about sharing their history, know their risks, recognize symptoms and problems, understand the care they need, are aware of the resources available to help them, and can access relevant care and services and (2) PCPs understand a survivor's history, know the survivor's risks, recognize symptoms and problems, understand the care survivors need, are aware of the resources available to help them, and can access relevant care and services. Specific outcomes that are assessed include loss to followup; surveillance for long-term and late effects; concordance with guidelines; communication, transfer of information, and care coordination; morbidity and quality of life; mortality; and costs.

We also found that there are an infinite number of models of survivorship care, and we identified four differentiating factors across models: (1) the inclusion of survivorship expertise (whether via a specialized PCP or oncologist; an MD, NP/PA, or multidisciplinary team); (2) the role of the PCP (e.g., main provider of survivorship care, provides survivorship care under the guidance of survivorship expert, provides primary care with no particular attention to survivorship); (3) degree of access to academic/cancer center support for survivors and/or providers; and (4) delivery of consultative versus longitudinal care. In practice, what is seen in the literature (and experienced by survivors) are more often patterns of care that occur not by design but owing to circumstance.

For the purposes of this project, the term “resources” included long-term followup guidelines; educational materials; trainings; survivor care documents (e.g., survivorship care plans); survivorship care management processes (i.e., expedited routes of contact for consultation); and survivor supportive tools and services (e.g., support groups). We identified 41 resources freely available to both survivors and providers: 25 survivor-specific, 10 provider/researcher-specific, 6 for both. We also identified 15 guidelines. Resources are most helpful if they are easy to access, user-friendly, known to survivors and providers, from trustworthy sources, and valued by survivors and their families.

With the answers to these CQs as a foundation, we explored how models of care that include primary care, and resources, could be effective for adult survivors of childhood cancer. To begin to understand the relationships among the factors related to childhood cancer survivorship care, we developed and then refined our initial program theory. This program theory describes how, nested in the overall environment, survivor and provider characteristics and facilitators/barriers interact to produce intermediate and then final outcomes. As shown in Figure 2, the models of care and resources connect with these factors at their intersection, leading to survivors' and PCPs' ability to obtain/deliver appropriate care as the intermediate outcome, and survivors living longer and feeling better through high-value care as the final outcome.

Building on this program theory, to address the realist review KQs (KQs 1 and 2), we developed seven CMO hypotheses, four focused on the survivor intermediate outcome and three

focused on the provider intermediate outcome. Because the models of care that include primary care (KQ1) and resources (KQ2) were closely linked, we addressed these realist review KQs jointly.

For the CMO hypotheses for survivors, we hypothesized that four mechanisms would be associated with higher levels of the survivor intermediate outcome: (1) linking resources with information for the survivor him/herself and to share with their PCP, (2) identifying perceived/actually healthier survivors and having perceived/actually lower needs for survivorship-specific care, (3) connecting survivors' engagement in the health system with increased knowledge about survivorship care, and (4) suggesting that survivors with greater confidence in their PCPs would be more willing to transition their care. For the CMO hypotheses for PCPs, we hypothesized that three mechanisms would be associated with higher levels of the provider intermediate outcome: (1) linking resources for the PCP to information needed to guide survivorship care, (2) identifying the shared care model as a way to obtain the needed support from oncologists, and (3) suggesting that PCPs with more experience caring for childhood cancer survivors would have greater comfort and expertise in doing so.

As described in the intermediate outcomes, at the most basic level, the models of care that include primary care, and the resources, seek to provide information to survivors and/or PCPs to enable them to obtain/deliver appropriate care. Thus, it is unsurprising that the variables from our program theory that were seen most consistently in the literature include oncology versus primary care, survivor and provider knowledge, provider comfort treating childhood cancer survivors, communication and coordination between and among providers and survivors, and delivery/receipt of prevention and surveillance of late effects. In turn, these were the variables that played the most prominent role in our CMO hypotheses.

Notably, our discussion of the CMO hypotheses also describe why they may not be effective in achieving the desired outcomes. For example, we hypothesize that information from resources is a key mechanism for achieving the intermediate and final outcomes for both survivors and providers. However, for survivors, we also discuss the evidence regarding how the information may be both beneficial and harmful, how the information may be more effective for some populations than others (e.g., survivors with lower baseline knowledge may benefit more; female survivors may engage with resources more), and the challenges of delivering the appropriate “dose” of information to be effective. Similarly, for providers, we note that their lack of awareness of resources, the possibilities and problems inherent with information in the EHR, and, again, balancing the “dose” of information so that it is useful without requiring undue burden. In this way, our CMOs describe both how various mechanisms could be effective, as well as why they may not be.

In combination, the answers to the CQs, the program theory, and the CMO hypotheses provide valuable insights into the how and for whom, in what contexts, and via what mechanisms models of care that include primary care and resources could be effective for adult survivors of childhood cancer.

Strengths and Limitations

It is important to consider these findings in the context of the strengths and limitations of this realist review. First, we were charged with answering the question of how models of care that include primary care *could* be effective – not whether they *are* effective. While not the focus of this review, we did find evidence that oncology specialty care is more effective in providing survivorship care for childhood cancer survivors.^{7, 9, 10, 26, 27, 32, 49, 55, 58, 60, 71-73}

It is also worth noting that, while this realist review aimed to address “models of care,” as described in the CQs, specific models of care are not clearly delineated and are rarely purposefully selected. Rather, the literature generally only provides evidence regarding “patterns of care” (i.e., who got seen where and received what), not evaluations of formal models of care. This limitation of the literature further complicated our realist review, which would ideally focus on a “well-defined program.”⁷⁴ In summary, rather than conducting a realist review of one well-defined model of care, we faced the challenge of conducting a realist review of multiple ill-defined patterns of care.

Strengths of our literature review and abstraction approach include the investigation of a wide range of papers, in multiple contexts, internationally. For the development of the initial program theory, we focused on commentaries, editorials, and qualitative and mixed-methods papers that described intended operations and outcomes of models of care and resources. During refinement of the program theory, we focused on empiric studies that could inform our program theory revisions and CMO hypothesis development. We not only included studies examining models of care and resources for adult survivors of childhood cancer, but also for child/adolescent survivors, as well as adult survivors of adult cancer. While the data from other populations added insights informing our CMOS, the generalizability of these findings to adult survivors of childhood cancer requires further exploration. We did not restrict our literature review to studies in the U.S., although the applicability of studies from other countries with different health systems may be limited. Given the short time available to conduct the literature review and abstraction, we could not go through as many iterations of developing theories and conducting additional searches to explore these theories further, as has been done in some other realist reviews. Rather, we largely conducted the review and abstraction in one large effort and then prioritized which studies we focused on in developing our CMO hypotheses. Specifically, our CMOs were developed primarily based on the literature for adult/adolescent/childhood survivors of childhood cancer, although we drew on the literature from adult survivors of adult cancer where it added explanatory value. With realist reviews, the goal of the literature abstraction is to be informative and illustrative, rather than comprehensive and confirmatory. There is an infinite amount of literature that could be reviewed, and time inevitably constrains how much is done.

A final strength of our realist review is the multidisciplinary expertise and extensive experience of our research team and Stakeholders. The research team included expertise in childhood cancer survivorship clinical care, survivorship research, oncology care, primary care, health services research, qualitative methods, and systematic reviews. We also received valuable input from our Stakeholders. Eight individuals provided a range of expertise and experience in childhood cancer survivorship research, advocacy, and clinical care; one expert advised on realist review methods. We consulted with these Stakeholders at three timepoints during this process: at project initiation, after developing the first draft of the initial program theory, and after revising the program theory and developing the CMO hypotheses.

Gaps in the Literature and Future Directions

Several gaps in the literature are noteworthy and should inform future directions in research in this area. First, there is a lack of formal evaluations of models of care, particularly models that include primary care. As described above, the lack of a clear taxonomy of models of care contributes to this problem. In addition, data regarding final outcomes, particularly mortality, are sparse (e.g., are survivors who are more adherent to recommended surveillance more likely to

live longer?). Other more specific issues mentioned in the Results, such as the possible need to tailor resources to different groups (e.g., males vs. females), should also be explored.

During the conduct of this study, the world experienced the transforming effects of the COVID-19 global pandemic. This factor is reflected in our program theory variable list as “crisis events,” but the literature has not even begun to reflect how medical care in general, and cancer survivorship care in particular, may be changed. For example, where the use of telemedicine was relatively limited in 2019, it became commonplace – and in some cases dominant, in 2020.⁷⁵

As described by one of our Stakeholders, the pandemic further emphasized two questions related to this review: (1) who needs to be seen in specialty care and who can be followed in their own community; and (2) for those followed in the community, how can the knowledge that survivors and PCPs need to receive/deliver quality care be effectively transferred? These questions represent the crux of the issues that require further research.

Conclusion

In summary, care for adult survivors of childhood cancer is complex, and the appropriate models for delivering this care are unclear. In fact, the various models of care are not well-defined, and what is seen in the literature (and experienced by survivors) are more often patterns of care that occur not by design but owing to circumstance.

While there is evidence that suggests that care delivered in a specialty setting is superior, various barriers outlined in our program theory (e.g., availability of specialized care, distance to care) describe why it may not be universally available. Our charge in conducting this realist review was to describe for whom and under what circumstances models of care that include primary care could be effective for adult survivors of childhood cancer. A common theme across the CMO hypotheses developed as part of this realist review is that, if care is going to be delivered outside of the specialty setting, there has to be knowledge transfer to survivors and PCPs.

This realist review identified a number of ways this knowledge could be shared, including a range of resources (e.g., guidelines, SCPs) and contexts (e.g., survivor confidence in PCPs, shared care with oncologists). The resulting program theory and related hypotheses elucidate some of the key CMO relationships that could be associated with effective survivorship care models that include primary care. Further research is required to explore whether these CMO relationships can be effectively actualized.

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Abbreviations and Acronyms

Acronym	Definition
AHRQ	Agency for Healthcare Research and Quality
ASCO	American Society of Clinical Oncology
AYA	Adolescent/young adult
CASP	Critical Appraisal Skills Programme
CMO	context-mechanism-outcome hypotheses
COG	Children's Oncology Group
CQ	Contextual Question
EHR	Electronic Health Record
EPC	Evidence-based Practice Center
GP	General practitioner
KQ	Key Question
MD	Medical Doctor
NAM	National Academy of Medicine
NCI	National Cancer Institute
NP	Nurse Practitioner
PA	Physician's Assistant
PCP	Primary Care Provider
RAMESES	Realist And Meta-narrative Evidence Syntheses: Evolving Standards
RCT	Randomized controlled trial
SCP	Survivorship Care Plan
STAR	The Childhood Cancer Survivorship, Treatment, Access, and Research (STAR) Act
TOO	Task Order Officer