



## *Systematic Review Disposition of Comments Report*

**Project Title:** *Models of Care That Include Primary Care for Adult Survivors of Childhood Cancer: A Realist Review*

Draft report available for public comment from May 18, 2021, to June 18, 2021.

**Citation:** Snyder C, Yuan CT, Wilson RF, Smith K, Choi Y, Nathan PC, Zhang A, Robinson KA. Models of Care That Include Primary Care for Adult Survivors of Childhood Cancer: A Realist Review. Evidence Report. (Prepared by the Johns Hopkins University Evidence-based Practice Center under Contract No. 75Q80120D00003.) AHRQ Publication No. 22-EHC003. Rockville, MD: Agency for Healthcare Research and Quality; February 2022. DOI: [10.23970/AHRQEPREALISTMODELSOFCARE](https://doi.org/10.23970/AHRQEPREALISTMODELSOFCARE). Posted final reports are located on the Effective Health Care Program [search page](#).

### **Comments to Draft Report**

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Comments on draft reports and the authors' responses to the comments are posted for public viewing on the website approximately 3 months after the final report is published. Comments are not edited for spelling, grammar, or other content errors. Each comment is listed with the name and affiliation of the commentator if this information is provided. Commentators are not required to provide their names or affiliations in order to submit suggestions or comments.

This document includes the responses by the authors of the report to comments that were submitted for this draft report. The responses to comments in this disposition report are those of the authors, who are responsible for its contents, and do not necessarily represent the views of the Agency for Healthcare Research and Quality.

## Peer Reviewer, Technical Expert, and Public Comments and Author Response

	Commentator & Affiliation	Section	Comment	Response
1	Peer Reviewer #1	Introduction	In describing childhood cancer survivors in the first paragraph, the authors make a distinction between those diagnosed in childhood and those in adolescence. However, through the rest of the review, they mention childhood only. Would suggest that to avoid confusion, they just point out ages in the introduction and specify that this report is focused on adult survivors (ages 18 and older) of childhood cancer (0-18).	We appreciate the reviewer noting the possible confusion and have added text under “Purpose and Scope” to clarify that we included survivors diagnosed prior to age 21 and, for simplicity, use “childhood” cancer survivor throughout.
2	Peer Reviewer #1	Introduction	Would also then be helpful to clarify whether this is focused on those who have completed treatment or may still be in treatment, or in early surveillance. The transition language is important especially for those who complete treatment, while model of survivorship care may include those on and off treatment.	We have clarified under “Purpose and Scope” that we included studies focused on survivorship care, not cancer treatment.
3	Peer Reviewer #1	Introduction	Clarify early on who is in the category of primary care, both degree (MD, NP, PA) and specialty (internal medicine, family medicine). How about pediatrics? OBGYN?	Under “Purpose and Scope,” we defined the degrees and specialties included in our review.
4	Peer Reviewer #1	Methods	Seems appropriate	Thank you for your comment.
5	Peer Reviewer #1	Results	writing is a bit dense, especially for an audience who may not be as invested and/or knowledgeable of this field. Would suggest adding space, bullets, boxes/tables. Would also suggest separating out studies focusing specifically on adult survivors of childhood cancer from those in other populations (such as GYN cancer survivors). Also, where the focus is AYA that may include other populations. As commented earlier, describe when studies including patients on treatment or shortly off treatment (in surveillance)	We ensured all study discussions mentioned whether it was conducted in childhood cancer survivors or adult survivors of adult cancers. In all cases, we discuss studies in childhood cancer survivor populations prior to studies from adult survivors of adult cancers.
6	Peer Reviewer #1	Results	Consider including a greater discussion about the resources found	We have added details describing the resources to Appendix E: List of survivorship resources available to cancer survivors, their families, and their medical care providers.
7	Peer Reviewer #1	Discussion and Conclusions	would try to make the text a bit less dense to read, add space, bullets, etc.	We have added space and bullets in the Discussion to improve the readability.

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8	Peer Reviewer #1	Discussion and Conclusions	more distinct ways that this review informs research and clinical practice. As it stands now, not clear 'next steps' presented. This should be a review that can be cited for future efforts in this field.	We have added that the results are intended to inform decision making by the National Cancer Institute regarding investments in future research, as well as how the review can more generally inform future research and health system planning to promote improved care and outcomes. ("Introduction: Purpose and Scope" and "Discussion, Summary of Findings").
9	Peer Reviewer #1	Discussion and Conclusions	in the limitations, the authors state that this review was to demonstrate of what could be effective rather than what is effective. Suggest that this is highlighted early in the review.	We added text about realist reviews and the purpose of this review under "Purpose and Scope" to emphasize this point.
10	Peer Reviewer #1	Appendix	No comment	
11	Peer Reviewer #1	General	The team was quite thorough in its review and presented key findings, and should be lauded for their effort. I am not sure if my relative "underwhelm" is due to the lack of evidence in this field or the lack of clinical significance of the findings.	Thank you for your comment. We hope the expanded text regarding the purpose and implications of the report improves the reviewer's overall impression.
12	Peer Reviewer #1	General	If the project goals permit, would suggest that there are clear 'next steps' here to inform research and clinical care.	As noted above, we have added that the results are intended to inform decision making by the National Cancer Institute regarding investments in future research, as well as how the review can more generally inform future research and health system planning to promote improved care and outcomes. ("Introduction: Purpose and Scope" and "Discussion, Summary of Findings")
13	Peer Reviewer #1	General	Lastly, this is focused on cancer-specific survivorship care, not overall comprehensive care.	We added a sentence under "Purpose and Scope" to emphasize this point.
14	Peer Reviewer #2	Introduction	The background in the Introduction succinctly provides a compelling rationale for the need for the review -- i.e., progress in survival and growing population at risk of multi-morbidity.	Thank you for your comment.
15	Peer Reviewer #2	Methods	Inclusion and exclusion criteria as justifiable, logical and specifically stated. The search strategies involved extensive efforts to identify evidence relevant to the topic. The methods used to appraise the quality of the literature were scientifically rigorous.	Thank you for your comment.

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16	Peer Reviewer #2	Results	The contextual questions are highly relevant. Summary tables enhance the data presentation. The Refined Program Theory is excellent and encompasses the spectrum of clinical targets within models of care that can facilitate or obstruct access to quality survivorship care. I do not think that the authors have overlooked relevant studies.	Thank you for your comment.
17	Peer Reviewer #2	Results	Inclusion of evidence from non-U.S. health care systems is less relevant, but still useful in providing context for survivor or provider factors that may be useful targets for research.	Thank you for your comment.
18	Peer Reviewer #2	Discussion and Conclusions	I think that authors very clearly address limitations of the literature and acknowledge that the proposed context, mechanisms and outcomes COULD be associated with quality survivorship care and improved outcomes. The CMOs and Refined Program Theory outline clinically meaningful targets to future research.	Thank you for your comment.
19	Peer Reviewer #2	Appendix	No comment	
20	Peer Reviewer #2	General	The report synthesizes available literature from a realist review into a program theory that was subsequently refined through discussions with stakeholders. The evidence presented provides extremely clinically meaningful information to guide development of interventions to support communication of knowledge to survivors and providers that could improve outcomes for childhood cancer survivors. The target population and audience are explicitly defined and the key questions are appropriate and clearly stated.	Thank you for your comment.
21	Peer Reviewer #3	Evidence Summary	'Main points' – the four points largely simply describe what was done rather than what was found – if I was trying to use this highlighted box to quickly learn/inform practice, I wouldn't have learnt much.	We agree that summarizing specific results (Figure A and Tables A and B) in a bullet point is challenging. We reviewed and discussed these points again. Of note, these main points were recently highlighted in the EPC Program as exemplars of good main points and, as such, we have retained these points.

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22	Peer Reviewer #3	Introduction	<p>The aims and research questions are stated clearly enough, but whilst 'decision-makers' as an audience are identified, the Introduction could go much further in situating the review's goal more clearly within healthcare system planning. My understanding (which the authors allude to on p.17) is that, due to the multiple overlapping models in use, some conceptual clarity is being sought about the key ways in which these models are intended to achieve desired outcomes.</p> <p>The conceptualization could then be used to plan focused evaluative (maybe experimental) research to test these re-conceptualised models. Is my understanding correct? Clarification about this is central to the endeavour. Some of these points are mentioned in the Conclusion, but if correct then they need to be up-front and clear in the Introduction.</p> <p>I'm still left wondering (even with reference to the protocol) what the 'state of knowledge' at the outset is with the models of care. That decision-makers need to know 'what works and why' is repeatedly stated, but a sense of the likely challenges for different survivorship care models needs to be provided, both for the reader's understanding and the review team's efforts. For example, is more known about challenges at the level of the individual rather than providers?</p>	<p>The reviewer raises several important points. The topic of the report was conceptualized by the U.S. federal agency requesting the review, rather than the team. The first paragraph of the "Purpose and Scope of the Review" is based on the initial scope of work that accompanied the request for task order proposals. We have added text to clarify that the results of the review are intended to inform future funding investments by the National Cancer Institute.</p> <p>The reviewer also asks about the "state of knowledge" at the outset; however, a stated purpose of this report in the scope of work was to summarize this "current state of the science and assess future research needs," which we have also added to the "Purpose and Scope of the Review."</p> <p>Finally, a key point which we have sought to clarify throughout the report is that the models of care, for example as listed by the American Society of Clinical Oncology, do not reflect the reality of survivorship care. Thus, this report contributes a re-conceptualization based on the 4 dimensions we identified and defined to describe different models of survivorship care that include primary care.</p>



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23	Peer Reviewer #3	Methods	<p>Methods for determining 'rigor' and 'relevance' would benefit from being tightened-up, for example:</p> <p>Rigor:</p> <ul style="list-style-type: none"> <li>- are all of the questions relevant when considering the wide range of included sources (editorials, commentaries, etc)?</li> <li>- are additional questions required to appraise included quantitative studies (or quantitative aspects of mixed-methods studies)?</li> </ul> <p>Relevance ('inclusion criteria'):</p> <ul style="list-style-type: none"> <li>- Why limit consideration of relevance to adult survivors of childhood cancer? Why not adult childhood survivors of other serious childhood diseases?</li> <li>- "Are the findings generalizable to our program theory?" is a rather circular question – what are the criteria by which you made this judgment?</li> </ul>	<p>We have clarified several points in the Methods based on the reviewer's comment.</p> <p>First, we have changed "Determinations" to "Considerations" as it more appropriately conveys our approach.</p> <p>Second, throughout the report, including under "Refinement of Initial Program Theory: Selection of Documents," we changed "empiric" to "quantitative" to clarify that this part of the review did not include qualitative studies, editorials, commentaries, etc." Thus, we feel that the questions regarding rigor were appropriate.</p> <p>We have also added text to explain that we gave more weight to studies with greater relevance but did not exclude others. For example, we did not exclude studies of adult survivors of adult cancers. We clarified that the question related to the program theory relates to the <i>initial</i> program theory and an evaluation of whether the article addressed the relevant variables and their associations.</p>

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24	Peer Reviewer #3	Methods	<p>The analysis process (p.6) is rather vague and difficult to understand without any examples of how extracted data was presented and considered. It is possible to specify in much more detail how a realist analysis is conducted (e.g. see&lt;&lt;link 1&gt;&gt; - it would improve the report considerably if the authors could provide more detail about how they conducted their analysis.</p>	<p>Thank you for providing a reference regarding suggested changes to the description of our approach. While we read the recommended article, it was self-described as “methodologically novel as it was conducted in parallel with the timelines and methods of a wider research project so as to inform intervention development.” Thus, both its methods and its application were distinct from our realist review. It also used different terminology such as “conceptual platform” and “explanatory accounts.” Given the complexity of realist review terminology, we did not want to add new terms that might be more confusing than helpful.</p> <p>However, we have substantially expanded the description of our analytic methods to provide more details as relevant to our methods and purpose of informing future research investment.</p> <p>Additionally, we have added a figure to graphically display the process, and Appendix C provides the literature abstractions we used to inform refinements of our program theory and CMO hypothesis development.</p>
25	Peer Reviewer #3	Methods	<p>A graphic showing an overview of the review process (program theory development, searching for evidence, selection of sources, program theory refinement, intersections with stakeholders) would significantly help to clarify what was done.</p>	<p>A figure has been added to the Methods to give an overview of the review process.</p>

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26	Peer Reviewer #3	Methods	Whilst the RAMESES reporting standards checklist has been completed, it would be very beneficial for the authors to also complete and reflect on the RAMESES quality standards <<link 2>> This would help clarify the rationale and operationalization of a realist approach and could usefully be included as an appendix (as well providing additional detail that could be included in the Methods of the main report).	Thank you for your suggestion. We believe the review demonstrates understanding and application of realist philosophy and we have added details to increase the transparency of the reporting; we have identified and developed an initial program theory; we have defined the search processes, selection and appraisal of documents, and data extraction.
27	Peer Reviewer #3	Results	I found it very disconcerting to have no overview provided about the characteristics (e.g. source type, number of participants, research method, whether survivor of childhood or adult cancer, results of critical appraisal etc etc) of the 135 studies included for program theory refinement. An 'included studies characteristics' table is a fairly standard expectation for a systematic review, whatever the approach.	Thank you for this suggestion. We have added details about the studies in the Appendix.
28	Peer Reviewer #3	Results	Presentation of findings could be considerably improved by providing e.g. a boxed overview of how the findings are structured, otherwise the reader is left to bounce around between 'title' context, various mechanisms, and subthemes.	Thank you for your comment. Tables 2 and 3 provide an overview and summaries of the results.
29	Peer Reviewer #3	Results	Findings are organized by 'context' – broadly, resources and survivor characteristics. Why were these contexts chosen?	The contexts were not chosen <i>a priori</i> , but emerged from our evaluation of the literature.
30	Peer Reviewer #3	Results	p.17 on - There is good level of detail provided in the Findings, but being presented as consecutive summaries of individual studies these can become rather hard to read. This is a challenge in many reviews, but the authors could take further steps to SYNTHESISE the findings from individual studies so that the narrative of the argument (the refined CMO configurations) is developed and made clear. Too often it is left to the reader to weigh-up where the weight of evidence is, with the summation of each section being equivocal rather than judiciously conclusive. Note, this is not always the case, e.g. Context B summation p.21 presents a judicious interim conclusion.	We appreciate the suggestion and have gone through the CMO Results and tried to add summative statements where supported by the literature and where not already present.



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31	Peer Reviewer #3	Results	Re: presentation of findings, whilst the Carrieri et al protocol is cited, the now-published main report could be instructive to the researchers re: presentation <<link 3>> , e.g. consider including a table of included studies, a summary of all CMO configurations explored, explanatory statements of the program theories (rather than the descriptive lists provided in Fig 2)?	Thank you for your comment. We have noted a diversity of approaches in realist reviews and feel comfortable that our report accurately and effectively represents our methods and results and conforms with the RAMESES standards.
32	Peer Reviewer #3	Discussion and Conclusions	Discussion/ Conclusion: p.29 Whilst it is recognised that the included literature reports a 'messy' bundle of models of care, it's unclear why it is claimed that the challenge faced was "conducting a realist review of multiple ill-defined patterns of care" when Table 1 shows ASCO's eight models of cancer survivorship care. It seems a significant missed opportunity to have not explored how the CMOs developed relate to these models or how the findings can inform the development of (or research required to inform development) these models. (Or, indeed, a rationale for focusing on developing a smaller number of models).	The reviewer raises an important question. A key point of this review is that the models of care as outlined by ASCO do not reflect what happens in practice or what is reported in the literature. Rather "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 have added text to clarify this point in the Results and Discussion.
33	Peer Reviewer #3	Discussion and Conclusions	The claim that "the lack of a clear taxonomy of models of care contributes to the [problem of few/no evaluations, particularly those that include primary care]" is bemusing given that a) realist reviews are often conducted because of its strengths in conceptual development (which can underpin a taxonomy); and b) the existing ASCO models of care (most of which include a primary care provider).	As noted above, a key issue is that the models of care as outlined by ASCO do not necessarily reflect what happens in practice, and few studies exist that formally evaluate different models of care. We have added text in the Results and Discussion to emphasize this point. We also highlight how this realist review identified four dimensions that can be used to define models of care as actually seen in practice and in the literature.
34	Peer Reviewer #3	Discussion and Conclusions	p.29 – the implication of the description of how 'empiric' studies were used to refine program theory appears to place qualitative and mixed-methods study in a 'non-empiric' category. Is this what is meant? The rationale for including different study types at different stages of the review should be clearly stated much earlier in the report (in Methods).	We appreciate your comment and have clarified in the Methods that we used opinion pieces, editorials, commentaries, and qualitative and mixed-methods studies for initial program theory development and focused on quantitative studies (including the quantitative aspects of mixed-methods studies) for program theory refinement.

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35	Peer Reviewer #3	Discussion and Conclusions	p.29 The observation that “there is an infinite amount of literature... and time inevitably constrains how much is done” may be true at a practical level, but is insufficient justification in a scientific report – in a complex area of inquiry, how were decisions made about what was and wasn’t focused on? Why should a reader of the report put their trust in the report’s findings, not as the final word on a very complex area, but in the stated aspects?	We agree that this point from the paragraph’s concluding sentence goes without saying, so we have deleted it. The paragraph itself clearly describes the Strengths and Limitations of our literature review approach to allow the reader to evaluate the findings in the context of our methods.
36	Peer Reviewer #3	Discussion and Conclusions	The review is probably a useful source about cancer survivorship models of care, but ultimately the conclusions seem rather pedestrian (e.g. individualized tailoring is beneficial, males and females engage in different ways with care planning, survivors who are already engaged with health care will benefit more from care plans, passive distribution of guidelines is unlikely to improve guideline-adherence). Is the strength of the review in bringing together these (perhaps unremarkable) patient, provider and health system understandings in one place so that decision-making about future cancer survivor model of care research and health system planning can be informed in a more rounded (less partial) way? The review’s distinct contribution to the literature and decision-making needs to be stated much more clearly.	We agree with the reviewer that it would be helpful to highlight how this realist review has investigated and critically evaluated the literature related to models of survivorship care for adult survivors of childhood cancer and the important role it can play in informing future research and health system planning. We have added a sentence under “Summary of Findings” to make this point.
37	Peer Reviewer #3	Appendix	An example data extraction table as an appendix would help clarify the way in which information/data was extracted and thus subsequently organized for analysis.	A set of example data extraction tables has been added to the appendices as Appendix B. These tables include Study Characteristics, Variables and finding, and Data assessment. Appendix C provides the data summaries we engaged with to refine our program theory and develop our CMO hypotheses.
38	Peer Reviewer #3	Discussion and Conclusions	All the steps of a realist review are documented, but this is largely about saying what was done and how the program theory changed rather than moving beyond description towards explanatory insights. I didn’t get a sense that the research team had really harnessed the potential of realist research to go beyond more conventional approaches.	Thank you for acknowledging we went through all of the steps of a realist review. As with all realist reviews, we could have gone even deeper given more time.

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39	Peer Reviewer #4	Introduction	Gaps are addressed, though the section could have benefitted from more description; did not get an entirely clear sense of how this study differs from a systematic review though it is implied that this is because there is a lack of formal models of care to evaluate.	To address this comment, we have added several sentences under "Purpose and Scope" regarding how a realist review differs from a systematic review.
40	Peer Reviewer #4	Methods	IN describing the realist synthesis approach, and in reading the report, it is confusing because the writer refers to "program theories" for individual interventions, and yet the product is also referred to as a program theory; can this wording be distinguished so that the 2 circumstances are distinct? Confusing to follow (e.g. page 14 of 47, lines 19 and 24, and then page 15 of 47, first paragraph---are the program theories the subject of study or is the program theory the product being developed? Is the wording intended to be the same? Are the conceptual models being studied instead of program theories??)	We appreciate this reviewer pointing out this area of confusion and have substantially revised the relevant sections to improve clarity.
41	Peer Reviewer #4	Methods	For stakeholder engagement (page 14 of 47, lines 45/46), were patients included as stakeholders? Caregivers and patient advocates are mentioned, but not patients themselves??	We did not have a patient stakeholder. We engaged patient advocates to represent the perspectives of patients.
42	Peer Reviewer #4	Methods	There is a typographical error, page 14 of 47, line 50, should read "throughout the project..." (missing a "the").	This has been corrected.
43	Peer Reviewer #4	Results	This is a very lengthy section and more figures here I think would be helpful. Subthemes did not seem to this reviewer to be represented in the final program theory figure?? This section was very detailed however and adequately addressed the questions addressed.	Thank you for your comment. We believe the subthemes are represented in the refined program theory. We agree this section is long, but details have been summarized in tables 2 and 3.
44	Peer Reviewer #4	Discussion and Conclusions	See comment above regarding use of the word "infinite", seems distracting and not perceived as being evidence based (page 37 of 47, line 25)	We have replaced "infinite" with "many" throughout.
45	Peer Reviewer #4	Discussion and Conclusions	Implications of discussions are adequate and limitations are discussed (namely the lack of studies evaluating a formal model of care), these are quite significant to address.	Thank you for your comment.
46	Peer Reviewer #4	General	This reviewer does believe that the topic and report is very important and meaningful. Though the overall population of adult survivors of childhood cancer survivors is small, the potential for serious late and long-term morbidity and mortality in this population is significant; many of these are potentially modifiable. The challenge as outlined is how to most effectively care for this population.	Thank you for your comment.

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47	Peer Reviewer #4	General	The methodology and wording for this report was a bit confusing and this reviewer had to read and re-read the report to fully understand the aims and goals (this reviewer was unfamiliar with this methodology prior to doing this review and I suspect that many other readers may likewise be unfamiliar).	We appreciate the reviewer's point and hope the edits made in response to the other comments have improved the readability and understandability of the report.
48	Peer Reviewer #4	General	On page 8 of 47, line 9, suggest changing the word "infinite" to "many" or some other wording, as pragmatically there are not "infinite" models of survivorship care. While there may be a lot it is not "infinite and the choice of wording here seems distracting a bit	We have changed "infinite" to "many" throughout the report.
49	Peer Reviewer #5	Introduction	P8 line 10 – suggest not including the word infinite	Throughout the report we have changed "infinite" to "many".
50	Peer Reviewer #5	Introduction	P8 line 27 – I don't understand this sentence: "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." Models and resources seek to provide information?	Thank you. We have edited this sentence to improve clarity.
51	Peer Reviewer #5	Introduction	Table B hypotheses: shared care with oncologist should include PCP explicitly. Is this table geared toward the oncologist or PCP? Does the hypothesis posit that the oncologist does not benefit from PCP involvement? I feel like I perhaps missed the premise that the patient is transitioning from oncology to PCP care, though shared care is mentioned here.	We clarified at the end of the Evidence Summary Results that the CMO hypotheses are focused on how models of care that include primary care, and resources, could be effective in providing care to adult survivors of childhood cancer. We retitled Tables A and B, as well, both in the Evidence Summary and in the main report to clarify the focus of the CMO hypotheses and specify PCP.

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52	Peer Reviewer #5	Introduction	<p>The motivation for the review sometimes eluded me. There seems to be a foregone conclusion that childhood cancer survivors are best served by involvement of PCPs, which I personally believe is likely true, but it is not really demonstrated in the intro or in the review. There also is an assumption that typically survivors are seen most intensively by oncologists and then in some way handed off or handed back to PCPs. The actual patterns of care are not described, nor how they are potentially lacking, nor how specific elements of care will contribute to the outcomes of better health and longer survival. (For example, should we expect the burden of late effects to be diminished if the appropriate care were received? To what extent are these late effects really mitigated by improved care? Where are the specific gaps in care that could be ameliorated with more PCP involvement?) Also, unlike survivors of adult cancers, survivors of childhood cancers transition not just from oncology to primary care (perhaps) but also from pediatrics to adult care, and this extra challenge (or at least component of care) is not addressed. I think the intro could be bolstered by at least minimally describing the care childhood cancer survivors receive, positing that including primary care is likely beneficial (if not necessary from an oncology shortage perspective), and that this review is an exploration of how pediatric oncology programs can integrate primary care to improve guideline adherence and improve health outcomes.</p>	<p>The reviewer has raised several important points, which we have tried to clarify in the report. First, we now clearly state under “Purpose and Scope” that the assigned topic of the review was to focus on models that included primary care. Second, our purpose was not to explore whether models of survivorship care that include primary care are more or less effective than models of care that do not include primary care, but to describe for whom and in what circumstances models of survivorship care that include primary care could be effective. Thus, it was also not within the scope of the review to assess current patterns of care or evidence of their association with outcomes. Our topic was more theoretical and focused on how models of survivorship care that include primary care could be effective. We do however note in the Discussion the lack of evidence regarding final outcomes, such as mortality to inform our theories.</p> <p>The important issues regarding care transitions are addressed in a separate EPC report as part of this National Cancer Institute-funded series.</p>
53	Peer Reviewer #5	Methods	<p>The key questions are appropriate and explicitly stated. Terms are very well spelled out throughout. Although I do not see “mid-range theory” defined. Methods seems appropriate and transparent.</p>	<p>Thank you for your comment. We have added a definition of mid-range theory under “Development of Initial Program Theory.”</p>
54	Peer Reviewer #5	Results	<p>Results page 8 – I can’t follow how the 62 included for initial program theory development were reduced to 34 records eligible from initial search. Were the remainders in the 322 already?</p>	<p>We have made revisions to the figure to make this transition clear.</p>

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55	Peer Reviewer #5	Results	P9 line 12: At face value, quality of life and morbidity may overlap. It may be too late to suggest aligning this with Nekhlyudov et al.'s framework for evaluating the quality of care for cancer survivors. There are many areas of overlap, but emergency department use and hospitalization could be included here, along with other thoughtful adaptations	Thank you for your comment on Contextual Question 1. The list was derived from multiple stakeholder meetings. We do not want to revise this list now as it is a reporting of discussions with stakeholders.
56	Peer Reviewer #5	Results	Page 14 model – This is a well-described model and reasonable to follow these components and connections. The provider side implicitly describes oncology (e.g. “return survivor to informed PCP”) and should maybe be labeled “oncology provider” or “cancer health care system” – to avoid the appearance that PCPs are not providers – and to clarify that the framework appears to be about connecting the treating institution to ongoing care. As another example, there is no inclusion of connection with the oncology team, which could be important for some survivors	We appreciate the reviewer raising this point as we intended for the provider side to reflect both oncology and primary care (e.g., “Willingness to transition/accept care for cancer survivors”). We have carefully reviewed the model and made edits to ensure that the provider side is not oncology-specific
57	Peer Reviewer #5	Results	Throughout the results, this framework anchoring at the treating institution is present, with connection to PCP as a desired outcome. Perhaps this should simply be acknowledged up front	We believe that the edits made to the “Purpose and Scope” section have clarified that connection to the primary care provider is the outcome of interest in the realist review (without assessing whether it is desirable).
58	Peer Reviewer #5	Results	Results page 14 and 15. Social determinants of health are included in the model for survivors and providers, contributing to barriers to care and as a match between providers and survivors that could enhance care. I wonder if some of these characteristics should appear in the provider health practices box as systemic approaches to reduce disparities in care.	While the reviewer makes an interesting point, it did not emerge from the literature we reviewed, and we have opted not to add it to the model.
59	Peer Reviewer #5	Results	The literature review is very clearly presented in CMO framework, and the subthemes are interesting and appropriate	Thank you for your comment.
60	Peer Reviewer #5	Results	Page 20 The resource “dose” as a theme is really a hypothesis that has not been tested. It has reasonable face value but there is no evidence shown	All of the CMOs and subthemes are, by definition, hypotheses. However, we have added text to note the need for confirmatory studies.
61	Peer Reviewer #5	Results	Page 20 line 50. Non-attenders rating GP-affiliated care higher than attenders is a little confusing as written and may not inform this section on how healthier survivors are less likely to use care.	We have clarified that because non-attenders were also less likely to report late effects than attenders of followup, we believe this finding supports the hypothesis that models of care that include primary care could be effective for survivors who are healthier.

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	Commentator & Affiliation	Section	Comment	Response
62	Peer Reviewer #5	Discussion and Conclusions	The finding that survivors and parents vastly prefer specialist care is telling, and it should be investigated whether the solution is improving trust in PCP or facilitating more specialist involvement. With generalists wanting more oncology involvement, and oncologist-based care found more effective (page 28, though it's not clear what the comparator is), it is not entirely convincing that what we need is more informed PCPs. To be clear, that may be a reasonable conclusion, or a necessary one given resources, but it is not obvious from this literature review. The conclusion is the spot in this review where this is spelled out most clearly.	The reviewer identifies an important need for clarification. The assigned topic of the realist review was to explore how models of care that include primary care could be effective for adult survivors of childhood cancer, not whether they are preferable to oncologist-focused survivorship models. We have now added text in the Introduction under "Purpose and Scope" to make this point upfront.
63	Peer Reviewer #5	General	Tables and figures are useful and clarifying, and they align with the conceptual model. The report is clinically meaningful, with the caveats mentioned below in the introduction about the motivation of studying this issue.	Thank you for your comment.
64	Peer Reviewer #6	Introduction	The introduction is excellent. It provides a concise summary of the literature and explicitly states the two aims of the realist review.	Thank you for your comment.
65	Peer Reviewer #6	Methods	The key questions encompass the main issues regarding models of care and resources. The KQs are relevant to both clinicians/program developers as well as to the multiple stakeholders interested in this area, including patients and insurers.	Thank you for your comment.
66	Peer Reviewer #6	Results	This is the section where the review really shined. All too often, reviews are a dry repetition of studies. The authors of this review demonstrated the ability to move beyond a simple recitation and provide a synthesis of the literature leading to Figure 2. The subthemes will also serve as a template for future research.	Thank you for noting your appreciation of our synthesis of the literature and the implications of our results for future research.
67	Peer Reviewer #6	Discussion and Conclusions	The discussion is succinct, yet provocative in calling for future research to improve the care delivery systems for childhood cancer survivors	Thank you for your comment.
68	Peer Reviewer #6	General	This is an outstanding synthesis of an area of care that, while needed, has lacked rigorous evaluations. In particular, Figure 2 is an important distillation of the key factors and relationships associated with the primary models of care. This figure will serve as a template for future studies to both develop/evaluate models of care but also to better understand multi-level factors.	Thank you for your comment. We appreciate your strong endorsement of the report's value.



	Commentator & Affiliation	Section	Comment	Response
69	Peer Reviewer #7	General	This was slightly laborious to delve into, particularly since I had some issues with the premise as noted above and there is a lot of term-defining and process-defining to start, but the figure and results were a really useful summation of the literature that felt complete and well-informed from stakeholders. This is a clearly written, thoughtfully executed review of an important issue, with transparent methods, a compelling framework, and interesting and complete results. Because studies generally don't inform long-term health outcomes or (usually) important preventive elements of care, there is a limit to what can be learned here about the value of primary care involvement, but that is not the fault of the review.	Thank you for your comment. In response to other comments, we hope we have clarified the premise of the report. We are pleased that our report reflects a useful and stakeholder-informed summation of the literature. We also appreciate your assessment of our clear writing and transparent methods.
70	Peer Reviewer #7	Introduction	The rationale for the review, objective and focus of the review are clearly stated. The target population is clearly defined. It would be useful to clarify who the intended audience of the review is for.	Thank you for your comment. In the "Purpose and Scope" section, we have clarified that the results of the review are intended to inform future funding investments by the National Cancer Institute.
71	Peer Reviewer #7	Methods	The key questions are appropriate and explicitly stated. The process and judgements around inclusion and exclusion of studies are clearly stated and there is a PARISMA diagram. The search strategies are stated and are appropriate for a realist review. The methods are clearly reported.	Thank you for your comment.
72	Peer Reviewer #7	Results	I think this was a challenging review to carry out as there is no formal, well defined model of care with associated documentation from which to identify a set of programme theories. Instead, the authors highlight that care is a matter of happenstance rather than planning, which is an important finding in itself. The authors have made a focused effort to understand the different patterns of care that occur and to identify how context shapes the mechanisms through which the care produced outcomes. They provide some interesting findings and tried hard to think in terms of and express the findings in CMO configurations.	Thank you for your comment.
73	Peer Reviewer #7	Results	I found Figure 2, p14 a little difficult to understand – it is more a representation of the different factors that might affect care, rather than a programme theory	We have seen a range of terminology in the field (e.g., program theories, conceptual platforms) and variations in what the terms represent. We have opted to retain use of program theory to avoid creating confusion.

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74	Peer Reviewer #7	Results	I found the table of CMOs easier to understand and very useful. I feel these could be strengthened further by providing more narrative around each programme theory, to explain how the c's, m's and o's interact.	While we appreciate the reviewer's comment, we feel that the Results section is already quite extensive and that the value of further elaboration would be offset by additional burden for the reader. In many ways, the Discussion section serves the purpose of the reviewer's suggestion.
75	Peer Reviewer #7	Results	The authors have tried hard to test and refine these and organize evidence under each programme theory. Perhaps a limitation is that there is still a lot of thinking in terms of 'variables' and much of the testing of theories seems to rest on the use of quantitative studies. This may reflect the dearth of qualitative studies that can shed light on mechanisms in this field.	We agree that further investigation of these issues using both quantitative and qualitative methods would be useful and have added this point to the Discussion.
76	Peer Reviewer #7	Discussion and Conclusions	These are clear	Thank you for your comment.
77	Peer Reviewer #7	Appendix	No Comments	
78	Peer Reviewer #7	General	Yes the report is clinically meaningful with an appropriate number of tables and figures	Thank you for your comment.
79	Steve Wosahla Children's Cancer Cause	General	<p>We believe the report gives minimal attention to the unique role of the guidelines as the current standard of care. Further education and awareness efforts to achieve greater uptake and utilization of the COG guidelines is needed.</p> <p>According to the report, a survey of US general internal medicine and family practice providers found that 93 percent reported never using the COG late effects guidelines and only 40 percent of providers were aware of the guidelines. However, another study reported substantial improvements in adherence to the COG guidelines between 2003 and 2016 and speculated that physician awareness of COG guidelines may be growing.</p> <p>Regardless of increased uptake, the current COG guidelines are lengthy and complex and would benefit from further study about barriers and whether a shorter version is needed for easy primary care use. The final report should recommend this action.</p>	While we appreciate this perspective, we have included discussion of guidelines consistent with what we found in the literature. Their role in providing information to survivors and their providers is addressed as appropriate in the context-mechanism-outcome hypotheses. We have also noted that the full potential of guidelines and other resources have yet to be fully realized and describe relevant sub-themes based on the literature.

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	Commentator & Affiliation	Section	Comment	Response
80	Steve Wosahla Children's Cancer Cause	General	<p>The report notes that 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. CCC agrees with this conclusion; however, multiple steps are necessary to achieve this goal.</p> <ul style="list-style-type: none"> <li>o First, a summary of care and survivorship care plan (SCP) must be delivered to both patients and families and primary care providers. The optimal time for delivery is likely at the first long-term follow-up visit, so that ample time can be offered to review the document and answer questions. Both items should conform with Children's Oncology Group guidelines.</li> <li>o Finally, multiple delivery mechanisms and formats are needed to address geographic disparities, survivor population differences, and provider capacity. A printout of the SCP is helpful during the visit as a tangible resource that can be explained point by point. But the SCP must also be documented in the medical record, sent to the PCP, and sent to the survivor. Survivors need an online version of the SCP, preferably one that can be viewed on a handheld device or from home (if they have a computer) and shared easily with other health care providers. There are multiple tools that assist with this, including Survivor Link and Passport for Care. New tools need to be specifically adapted for use in medically underserved, lower-income communities with limited access to WIFI, home computers, and smart phones.</li> </ul>	While we appreciate this perspective, the reviewer's recommendations go beyond what we found in the literature.
81	Steve Wosahla Children's Cancer Cause	General	The report found that developing and delivering a SCP is a uniformly good practice. However, use of the SCP by primary care providers, even when it is in the medical record, can be limited. The report found that the timing and type of intervention (tailored and specific) was relevant. Future demonstrations should include and evaluate how best to flag SCPs for providers considering mechanisms such as quality measures, use of electronic medical records, or other mechanisms. The report recommendations should highlight this issue.	We appreciate this comment and believe the key points consistent with the literature have already been included in the report.
82	Steve Wosahla Children's Cancer Cause	General	The report found, and we agree, that further research is required to evaluate the above knowledge transfer mediums and contexts for effective actualization.	Thank you for your comment.

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	<b>Commentator &amp; Affiliation</b>	<b>Section</b>	<b>Comment</b>	<b>Response</b>
<b>83</b>	Steve Wosahla Children's Cancer Cause	General	The report found improved knowledge for survivors "who were given a passport card describing diagnosis, treatment, risks, and recommended follow up. . . were more likely to demonstrate improved knowledge versus survivors without a passport." We feel the report should include further information about successful passport models, including mobile access to the personalized SCP.	Thank you for your comment. The report includes information on this topic based on what we found in the literature.
<b>84</b>	Steve Wosahla Children's Cancer Cause	General	The report supports a major goal of CCC – the authorization of a Center for Medicare and Medicaid (CMMI) demonstration program to develop standards of care for survivors of childhood cancer with a focus on scalable models based on the COG guidelines across the United States. Attached is a CCC proposal for such a demonstration entitled the Child and Adolescent Cancer Survivorship Transition (CAST) Model. We recommend that the report include the proposal as a strategy to overcome barriers to survivorship care.	While it is not appropriate to include the CCC proposal as part of the report, to the extent the findings of the report support the CCC's approach, CCC is welcome to reference the report.
<b>85</b>	Steve Wosahla Children's Cancer Cause	General	In conclusion, the report notes that real world evidence documenting the quality of the care adult survivors of childhood cancer receive is limited. The report also found a lack of formal evaluations of outcome data for the models of care, particularly mortality. Specifically, the report states, "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." CCC believes that real world evidence is the next step in improving survivorship care for childhood cancer survivors.	Thank you for your comment.

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86	Steve Wosahla Children's Cancer Cause	General	Not surprisingly, the report found few major studies focused on childhood cancer survivors and were forced to supplement with evidence from adult survivors of adult-onset cancers. The final draft should indicate where a study included survivors who were diagnosed with cancer as a child vs. diagnosed as an adult. Additionally, the final draft should note where studies include both pediatric and adult-onset cancers (if the childhood cancer survivor population composes <20% of the overall study population). The report should address this issue both broadly and study by study. Reported studies regarding childhood cancer survivors are lacking, however survivorship needs vary based on cancer onset by age. Thus, the report should clearly identify the study cohorts since the objective is to describe access to care barriers experienced by childhood cancer survivors.	We have made revisions to the results that clarify the population.
87	Susan Weiner Children's Cancer Cause	General	The AHRQ Report review effectively organizes literature on "an infinite number of models of survivorship care." It offers key summaries of data that can guide a framework for care, healthcare plans, providers and survivors to help improve the quality of survivors follow-up care. The Report's narrative is likely to be more accessible to stakeholders, however, if the format was simplified and key points were highlighted.	We feel that the Evidence Summary provides a more simplified format with key points highlighted. The text of the report has been revised, based on comments from this and other reviewers, with a view to improving clarity and transparency.
88	Susan Weiner Children's Cancer Cause	General	Evidence is strong that SCPs can be effective in educating survivors and even prompting their PCPs to take additional steps to care for survivors. However, many PCPs seem to be unaware of SCPs and guidelines. This finding indicates that a clear remedy is needed for professional training through a standard mechanism, e.g., CME course, medical school curriculum or fellowship training. The AHRQ fails to note this research gap, which is one of the main recommendations of the IOM report (#4).	The report summarizes the findings based on the literature. We have been careful not to over-interpret the findings by making statements that go beyond the literature.

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89	Susan Weiner Children's Cancer Cause	General	<p>The influence of cost and reimbursement for care warrants more salient attention in the AHRQ Report. The Report cites cost as a variable reported in many studies. Stakeholders expressed particular concern about cost of care. However, the Report offers no further analysis on this issue. As an example, the Report notes that oncology specialty care delivers superior care and cites that its availability is likely limited by distance to care settings. Cost is probably a more important variable affecting survivors' willingness to seek care. In addition, expenses associated with the delivery of quality care need to be delineated, for example costs associated with resources and coverage for transitional episodes for the transfer of information between survivor and care provider as well as ongoing episodes of follow-up care. The AHRQ Report could have made these point with more extensive analysis of cited studies.</p> <p>Reimbursement for care delivery is clearly an important factor in how care is shared between oncology providers and PCPs. Survivors clearly favor follow-up care by oncology providers, and PCPs favor some type of collaborative relationship with oncology care providers. The Report notes the tension between "providing sufficient 'dose' of information to be effective and being realistic regarding the time and effort providers can invest" (p.25), clearly an important reimbursement factor. Highlighting the issues of reimbursement between oncology and primary care in the report could provide a solid rationale that could stimulate others to conduct research on care costs to inform the creation of economically valid care models.</p>	<p>Thank you for your comment. In fact, we note in the second paragraph under "Refinement of Initial Program Theory" that cost was relatively infrequently addressed in the studies. Thus, we are limited in what we can say regarding these points.</p> <p>However, as noted, we do discuss associated issues as part of our context-mechanism-outcome hypotheses (e.g., balancing the "dose" of information to be effective with the time and effort required).</p> <p>We have also added a sentence in "Gaps in the Literature and Future Directions" regarding cost and other variables that were found less frequently in the literature.</p>

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90	Susan Weiner Children's Cancer Cause	General	The AHRQ report might have organized the diffuse literature differently with a different hypothesis. For example, if the goal had been what models might deliver quality care for adult survivors of childhood cancer, the hypothesis might have been what is the evidence that might structure a successful transition to adult care providers. The severity of long term and late effects and the nature of the chronic disease sequelae might have been used to stratify and understand how care was delivered in study reports. Stratification might suggest a gradient of survivorship care services through a continuum of service intensive care settings, either in clinics or communities. Such an approach might increase the cancer survivors' confidence in PCPs, tailor oncology/PCP interactions to survivors' needs, enable PCPs to take better care for adult survivors with less morbidity, and ensure that adults with chronic conditions move from pediatric settings to adult specialists in those chronic ailments, e.g., cardiologist, endocrinologists, gynecologists.	Thank you for your comment. Another EPC was charged with reviewing evidence about the transition to adult care.  We organized the report based on the assigned topic. Nevertheless, our context-mechanism-outcome hypotheses did at a very basic level stratify by noting that evidence suggests that models of care that include primary care may be effective for "Survivors who perceive themselves to be, or are actually, healthier"

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