An investigation of cultural influences in survivors of paediatric cancer: a systematic review protocol

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ABSTRACT

Introduction Paediatric cancer affects children and families from diverse backgrounds. However, there is a limited understanding of how diversity/cultural factors play a role, especially in survivorship. This protocol outlines a systematic review on the cultural influences in survivors of childhood cancer.

Methods and analysis This protocol is reported based on the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA) guidelines checklist and is registered with PROSPERO. EMBASE, MEDLINE and PsycINFO are searched. Eligibility criteria include original research studies published in English, and an assessment of culture on survivors of childhood cancer. Search terms are developed with a medical librarian. Primary objective will be to describe culture (ethnic and population groups, migration status, acculturation, cultural characteristics) in survivors of paediatric cancer and study characteristics and methods. Secondary objective will be to identify the role of culture in outcomes of survivors of paediatric cancer. Data extraction will include participant characteristics such as the number of participants and/or controls, sex, age at diagnosis. Extraction will also include analytical approaches, type of cultural variables (predictor, moderator, mediator, outcome) and effect measures.

Ethics and dissemination Ethical approval was not required for this systematic review. Results from this systematic review will be disseminated in line with PRISMA guidelines through peer-reviewed publications and conference presentations. Findings will also be shared with our target communities, including survivors of childhood cancer and their families, through the creation of lay summaries and/or educational workshops in the community. Knowledge gathered from this review may help to identify gaps in knowledge and directions for future research. They may also inform the development of clinical recommendations for healthcare providers of survivors of childhood cancer.

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INTRODUCTION

With the advancement of paediatric cancer treatment, there is a rapidly growing population of long-term survivors of childhood cancer. This calls to attention the need to better understand the experience of all survivors and ways to inform their long-term follow-up care. Unfortunately, research on the cancer experience of survivors of childhood cancer with consideration of their cultural context is quite limited. In a literature review conducted by Gray et al, the authors examined cultural factors in clinical care and family management for children and their families in various aspects of their cancer experience, ranging from diagnosis, active treatment, to cure/palliative care.1 The authors concluded from their review that the ways in which cultural factors influence survivorship is poorly understood. Existing studies conducted on the role of culture in the care of survivors have focused on specific ethnic and population groups, including those from Korea2 3 and Taiwan,4 as well as Latino youths in the United States5, regarding their health-related quality of life. These studies uniquely offer a perspective of the lived experience of survivors from selected cultural groups. However, given the scarcity of existing research on this topic, little is known about how culture affects survivorship across a wider range of cultural groups and geographical regions. For example, for survivors of paediatric cancer and their families that live in Canada, issues of acculturation and migration status may be of relevance to their cancer experience and follow-up care. As it stands,
research on the influence of culture on survivors of childhood cancer across cultural groups and countries has largely been neglected.

Broadly, the notion of culture can refer to specific groups of individuals who exist in a shared context, including their general characteristics, ideas, beliefs, technology, habits or practices. As such, studies on culture can vary widely but may include considerations of race, ethnicity, and socioeconomic status (SES), among many others. The recently published psychosocial standards of care for childhood cancer highlighted that low income or education is a risk factor that contributes to ongoing social and professional difficulties for survivors. Specifically, lower SES coupled with other contextual factors (eg, age at diagnosis) have been found to be associated with a greater degree of challenges in survivorship. In the same review, ethnic differences were also observed in the cancer experience of survivors. For instance, based on an ethnically diverse sample of adolescents and young adults survivors of paediatric cancer, Arpawong et al found that survivors who identify as Hispanic and spoke primarily English at home experienced lower post-traumatic growth than those that identified as Hispanic and spoke primarily Spanish at home, and those that did not identify as Hispanic.

Taken together, there is some evidence to suggest that cultural factors play a critical role in the late effects and quality of life for survivors of childhood cancer. However, there is a lack of a clear and complete understanding of how cultural factors fully influence paediatric survivorship. Further, to the best of our knowledge, a theoretical framework that accounts for the cultural context in paediatric survivorship currently does not exist. Prior research has largely focused on the effects of family resources (SES, income) and less is known regarding other cultural/ diversity factors, as well as their intersection, in paediatric oncology and especially for survivors. The majority of the work on survivors of childhood cancer are either based on Eastern countries or Hispanic immigrants to the United States. Gaining an understanding of the experience of survivor immigrants from other minority groups to North America may be fruitful to our understanding of how acculturation can impact families in their understanding and engagement in treatment, and how such experiences may vary across different ethnic groups in North America. On a global level, knowledge of how survivorship differs across different cultures and geographical regions can help to enhance the generalisability of our findings on and care for all survivors around the world.

Based on these gaps identified in the literature, there is a need to capture the full range of cultural factors that can potentially affect survivors. Broadly, a deeper understanding of the complex and dynamic process in which culture shapes children and families’ cancer experience is critical to provide optimal care. More specifically, knowledge of the experience of survivors from diverse backgrounds can help to determine key aspects of their cancer experience that can be impacted by their cultural context.

This knowledge can in turn inform healthcare providers in their delivery of culturally informed care to diverse populations. Accordingly, we endeavour to evaluate the evidence of cultural influences on survivors through a systematic review of the literature. On the basis of parsimony, we narrowed the scope of our review to focus on the study of cultural groups and their influences on the clinical, psychological, and social outcomes of survivors of paediatric cancer. Our conceptualisation of culture in this review is determined by ethnic and population groups, migration status, acculturation levels, and cultural characteristics (beliefs, attitudes, and behaviours). Our conceptualisation of survivors’ outcomes include their clinical outcomes (eg, treatment engagement and outcomes, late effects, follow-up care), psychological outcomes (eg, anxiety, depression), and social outcomes (eg, educational and vocational attainment).

**Study objectives**

The overarching review question is: How does culture influence survivors of paediatric cancer? To answer this question, specific objectives are outlined below.

Objective 1: To describe the studies that investigate cultural influences in survivors of paediatric cancer, including study characteristics (eg, conceptualisations, operationalisations) and current methods used to measure cultural factors in survivors of paediatric cancer (eg, study design, sample size) coming from a systematic review.

Objective 2: To identify the effects and/or interactions of cultural influence on the outcomes of survivors of paediatric cancer, including psychological, social and clinical outcomes.

**METHODS AND ANALYSIS**

This protocol is registered with PROSPERO (CRD42021324101) and reported in accordance with the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) statement. This systematic review started May 2021 with a preliminary search, pilot of the study selection process, and screening of search results against eligibility criteria. As of February 2022, no data extraction has been conducted. The PRISMA-P checklist was completed. Systematic review progress will be updated on PROSPERO, including any changes to the planned systematic review along with a rationale for the changes.

**Patient and public involvement**

This systematic review protocol was developed with patient and public involvement. Specifically, the introduction, objectives and methodology of this systematic review was developed with feedback provided by our programme’s patient partners. We will also collaborate with patient partners on the interpretation and dissemination of results.
Literature search/inclusion criteria

We will include original research studies that reported the association between cultural influences and outcomes (clinical, psychological, social) of survivors of childhood cancer. Studies must be published in English. Studies that included a wide range of ages and/or a variable time from diagnosis and treatment will be included if the mean age and/or time meet the aforementioned criteria. Studies will include quantitative study designs. Narrative review articles and grey literature such as conference and poster abstracts, dissertations and theses will be excluded. Original study authors will not be contacted directly.

Children, adolescents and young adults diagnosed with cancer between 0 and 21 years of age and who are at least 5 years from diagnosis and/or 2 years from the completion of therapy will be considered. Our definition of survivors of childhood cancer is consistent with the definition of survivors established from the Children’s Oncology Group guidelines. Studies in the context of health, paediatric, psychological, and social/cultural research will be considered. Studies conducted in any geographical locations will be included. No publication date filter was implemented to capture the full range of studies conducted on this subject.

Objectives

The primary objective is to describe culture in survivors of paediatric cancer. Culture in this review is defined as the study of cultural groups. Study characteristics will therefore include: ethnic and population groups, migration status, acculturation levels, and cultural norms (beliefs, attitudes and behaviours). Other culture-related measures used within our scope and conceptualisation will be considered. Type of cultural variables considered include predictor, control, moderator or mediator roles.

The secondary objective is to describe the effects and interactions of culture on clinical, psychological and social outcomes. Clinical outcome variables include (but is not limited to): late effects, follow-up care and treatment outcomes. Psychological and social outcomes will be characterised according to psychological and social categories. Specifically, psychological outcome variables include (but is not limited to): depression, anxiety and quality of life. Social outcome variables include (but is not limited to): peer relationships, family relationships, educational and vocational attainment.

Comparator (controls)

Comparison of intervention/exposure to a control group will be considered but not required. Cross-cultural comparisons, wherein a group of survivors from one country, ethnic or cultural group is compared with those of another country, ethnic or cultural group, will be considered but not required.

Information sources and search strategy

PRISMA guidelines for completion of systematic reviews will be followed. A literature search will be conducted using the following databases: EMBASE, MEDLINE and PsycINFO. Search terms will be developed with reference to established search filters and in collaboration with a medical librarian. A sample of search strategy for OVID Medline can be found in online supplemental information.

Study selection

All exported articles will be subjected to a two-step review process via COVIDENCE. In the first step, all available titles and abstracts will be screened by two independent raters to identify potentially eligible studies for inclusion. Disagreements will be resolved through consensus between these two raters. Inter-rater reliability will be calculated using the Kappa statistic. The identified studies based on the review of titles and abstracts will be retrieved in full and again, two independent reviewers will review the content of each full article for eligibility. Disagreements will again be resolved through discussion between raters. Inter-rater reliability will be calculated using the Kappa statistic.

Data extraction and management

Retrieved articles will be exported to COVIDENCE, a systematic review management tool to screen title/abstracts and full texts, and extract data. Data extraction will include the following data: study design type, number of participants, number of controls, sex, median age at diagnosis, age range at diagnosis, median age at evaluation, age range at evaluation, participation rate, median follow-up time, range of follow-up time, outcomes assessment method used and brief summary of study findings. In addition, analytical approach (eg, regression, tests of interaction between groups, correlations) will be extracted. Type of study cultural variable (predictor, moderator/mediator) and country in which the study was conducted will be extracted. Where possible, effect size and confidence intervals of the effect of predictor (culture) on outcome variables (clinical, psychological, social) will be extracted to determine the effect measure for my main outcome variables.

Quality assessment

Quality assessment will be conducted within each study by considering four primary criteria: (1) selection/subject bias; (2) instrumentation; (3) missing data and (4) reporting measurement outcomes. The quality of evidence across studies will also be evaluated using the Grading of Recommendations Assessment, Development and Evaluation framework.

Data analysis and synthesis

To address our first objective, studies will be summarised using descriptives based on the following criteria: conceptualisation of culturally relevant variables, operationalisation of culturally relevant variables (ie, measurement tool used); study design, sample size, characteristics of paediatric survivor group studied and reporter derived from...
the data extraction form described above. These findings will be described qualitatively using a narrative synthesis.

To address our second objective, all studies that explore culture and survivors of paediatric cancer will be evaluated to determine if they are associated with any clinical, psychological, and/or social outcomes. Data extraction will adhere to the clinical, psychological, and social categories as defined in this study. Data extracted on the effect measures will be summarised quantitatively. Currently, there is no childhood cancer-specific model that accounts for the influence of cultural factors on their outcomes. As such, evaluation of the associations between cultural variables and survivors’ outcomes will be guided by an existing cultural/acculturation framework (eg, see work by Ward and Geeraert).

ETHICS AND DISSEMINATION
This protocol outlines a systematic review focused on examining the cultural influences in survivors of paediatric cancer. This review does not require ethical approval. On completion of the systematic review, details of the final publication and/or preprints will be documented and displayed on the public record on PROSPERO. Findings will be shared with academic and clinical stakeholders through traditional academic outputs, including open-access journal publications and national and international academic conferences. Findings will be also shared with our target communities, including survivors of childhood cancer, in collaboration with patient partners, by creating lay summaries, educational workshops and/or online blogs.

Limitations
This systematic review protocol has limitations worth noting. Culture encompasses a wider range of factors that is not limited to the current variables examined. Other diversity factors such as religion, SES, sex, gender and ability level are also important factors to consider in the context of survivors’ outcomes. Future research on these latter factors can further contribute to our understanding of diversity/cultural factors in play for survivors of paediatric cancer. Another limitation of the current review is our inclusion of original research studies published in English only. This criteria precludes a review of studies conducted in other languages that may be fruitful to our research question and therefore limit the generalisability of our findings.

Implications
This review holds both research and clinical implications. First, the current review will advance our understanding of survivorship from diverse populations, with a focus on examining the influence of their cultural context/background on their outcomes. Outcomes of this work will in turn help to increase generalisability of research in paediatric oncology with diverse populations, as well as guide future studies in this area. This review may also inform the development and delivery of concrete and culturally specific clinical recommendations for survivors of childhood cancer which can in turn address health disparities among under-represented groups. Ultimately, a deeper understanding of the complex and dynamic process in which culture shapes children and families’ cancer experience is critical to provide optimal care.

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Contributors All authors contributed to the study conception and design, including the literature search strategy. The first draft of the protocol was written by SHJH. Subsequent revisions and edits were provided by FS. All authors read and approved the final manuscript.

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