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Mapping the concept, content and outcome of wilderness therapy for childhood cancer survivors: protocol for a scoping review

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ABSTRACT

Introduction Long-term childhood cancer survivors are at risk for frailty and have significant health-related issues in adulthood. Various health promotion interventions have been proposed to enhance quality of life including wilderness therapy, which applies the impact of nature on health in a therapeutic context. Previous studies have described positive outcomes linked with various wilderness-related therapies for cancer survivors. However, there is no clarity on the role these therapies play in childhood cancer. The current scoping review aims to systematically map the concept, content and outcome of wilderness therapy for childhood cancer survivors.

Methods and analysis This review will be guided by the Joanna Briggs Institute Reviewers' manual for scoping reviews. A systematic literature search using medical subject headings (MeSH) and text words related to wilderness therapy and childhood cancer survivors will be performed in EMBASE, ERIC, Medline, Psycinfo, CINAHL, Scopus, Web of Science, SPORTDiscus and Svemed+, Sociological Abstracts, supplemented by grey literature searches. Eligible quantitative and qualitative studies will be screened, included, assessed for quality and extracted for data by two reviewers independently. Results will be described in a narrative style, reported in extraction tables and diagrams, and where appropriate in themes and text.

Ethics and dissemination This study describes a protocol for a scoping review that will undertake secondary analysis of data already published in literature and is therefore exempt from medical ethical review. The scoping review will inform understanding of the benefits and risks of wilderness therapy for childhood cancer survivors, their families, practitioners, clinicians and researchers, and will help elucidate the steps necessary for building its evidence base going forward. Results will be published in a peer-reviewed scientific journal.

Strengths and limitation of this study

- ► To the best of our knowledge, no previous work has been carried out to systematically map and categorise the concept, content and outcome of wilderness therapy for childhood cancer survivors.
- ► This scoping review will provide insights on the benefits and risks of wilderness therapy for childhood cancer survivors and will inform the building of an evidence base going forward.
- ► This scoping review is of interest to a broad audience, including childhood cancer survivors, their families, practitioners, clinicians and researchers who have an interest in better understanding the role of wilderness therapy in childhood cancer.
- ► No meta-analysis or other statistical analysis will be performed in this review.

> Few places in this world are more dangerous than home. Fear not, therefore, to try the mountain-passes. They will kill care, save you from deadly apathy, set you free, and call forth every faculty into vigorous, enthusiastic action. Even the sick should try these so-called dangerous passes, because for every unfortunate they kill, they cure a thousand. (John Muir, The Mountains of California, The Century Co., New York, 1894)

INTRODUCTION

Worldwide, the incidence of childhood cancer is increasing. Recently published data report that the cancer incidence among children aged 0–14 years increased from 124.0 per million person-years in the 1980s to 140.6 in the period 2001–2010,[1] with an increase of about 0.6% per year.[2] In the USA alone, cancer incidence among 0–14 years-olds increased from 116 per million in 1975 to 160 per million in 2014.[3] Among adolescents and young adults, rates per million over this same period increased from 130 to 183 per million.[3] The most prevalent cancers in children are leukaemia, central nervous system tumours and lymphomas.[1] Due to advances in diagnosis and use of multimodal treatment, the percentage of children that survive cancer is also increasing. The current 5 year survival rate for childhood cancer exceeds 80%, although survival rates by cancer type

vary significantly[2–4] and global inequality in childhood cancer survival is still large. For example, age-standardised childhood survival for lymphoid leukaemia is 52.4% in Colombia and 91.6% in the German cancer registries.[4]

Given the growing population of childhood cancer survivors, increasing attention has been placed on health promotion interventions to improve the quality of life for children during and after cancer. Long-term childhood cancer survivors are at risk for frailty and lower quality of life and have significant disease and treatment-related health issues in adulthood.[5 6] At least 66% of cancer survivors between the ages of 5 and 19 years, and 88% of childhood cancer survivors between the ages of 40 and 49 years have one or more chronic diseases.[7] It has been reported that they have a higher risk for the development of stress-related mental disorders,[8 9] social, academic and vocational difficulties,[10 11] secondary cancers,[12 13] as well as increased risky health behaviours.[14–16] Sedentary behaviour and obesity are higher in childhood cancer survivors compared with siblings,[14–16] while rates of alcohol and tobacco use are comparable[14 17] or slightly reduced,[18] but problematic given underlying health risks related to cancer treatment. Furthermore, adolescent cancer survivors are at higher risk for depression and anxiety, social problems, problems with attention and learning, and decreased physical and mental health status compared with controls.[19–25] A systematic review reported that the majority of adolescent survivors are mentally healthy, but a significant subset report global psychological distress, problems with emotional functioning and post-traumatic stress.[21]

Various health promotion interventions, such as social skills development, physical activity, workbooks, education and web-based programmes may enhance quality of life for childhood cancer survivors and be helpful for their families.[26–28] Furthermore, interventions that increase social interaction are strongly recommended to decrease distress, decrease isolation and improve adjustment.[10 29] There is a renewed interest in interacting with nature as a health promotion strategy to increase quality of life.[30 31] Wilderness therapy is an example of the application of nature-human interaction in a therapeutic context. So far, there is no universally accepted definition of wilderness therapy. Adventure therapy is regarded the umbrella term under which a large variety of approaches appear such as wilderness therapy, nature therapy, recreation therapy, outdoor therapy or open-air therapy.[30] Russell defined wilderness therapy as an intervention that utilises outdoor adventure activities—such as primitive skills and reflection—to enhance personal and interpersonal growth.[31] Another definition by Davis-Berman and Berman describes wilderness therapy as a group treatment modality in mental healthcare that seeks to augment the restorative qualities of nature in combination with structured and intentional individual and group-based therapeutic work.[32] Fernee *et al*[33] proposed that wilderness therapy distinguishes itself from the larger group of wilderness experience programmes in that it encompasses elements specifically targeted toward the treatment of adolescent emotional, behavioural, psychological and/or substance use issues. They developed a wilderness therapy clinical model based on the milieu model of Russell and Farnum,[34] with core therapeutic elements being wilderness, the physical-self and psychosocial-self.[33]

Several reviews have reported on the beneficial health effects of nature-based programmes.[35–37] A narrative review of nature-based experiences for cancer survivors described positive effects of a number of outdoor activities, and concludes that being in nature supports quality of life, sense of belonging and self-esteem, as well as decreases state anxiety.[37] A systematic review and synthesis of qualitative studies on the role of nature in the lives of cancer patients and survivors reported that nature supported patients in navigating the clinical and personal consequences of cancer.[35] A recent scoping review aimed at identifying whether specific therapeutic factors of adventure therapy were more effective for any given (patient) population compared with other therapeutic interventions reported.[36] Little to no difference was found.[36] One congress abstract reported on a systematic review investigating the benefits of wilderness programmes for cancer survivors and patients.[38] (To our knowledge, the full systematic review article has not yet been published.) According to the abstract, the authors included systematic reviews and controlled studies to investigate the impact of wilderness therapy on health-related outcomes in cancer survivors (presumably adults) but did not include other designs or focus on childhood cancer survivors. None of the reviews described above have specifically addressed or investigated the role of wilderness therapy in childhood cancer survivors. The present study was therefore initiated to collect information on wilderness therapy for childhood cancer survivors. The results of this study will facilitate better understanding of the concept, content and outcome of wilderness therapy for childhood cancer survivors and will guide the development and optimisation of future wilderness programmes, identify knowledge and evidence gaps in the literature, and inform clinicians and researchers in the design of future studies on its efficacy.

A preliminary search for existing reviews on this topic was conducted in databases of the Joanna Briggs Institute, the Cochrane Database of Systematic Reviews, PubMed and CINAHL. No protocols for a similar review were found.

METHODS AND ANALYSIS

Aim and research questions

The aim of this review is to map the concept, content and outcome of wilderness therapy for childhood cancer survivors. Research questions were derived from the Population–Concept–Context (PCC) mnemonic recommended by the Joanna Briggs Institute.[39] The *population* is childhood cancer survivors; the *concept* is the underlying theoretical framework, content and benefits of

Box 1 Population–Concept–Context (PCC) mnemonic

Population
- Childhood cancer survivors.

Concept
- Wilderness-related therapies, including: adventure therapy, recreation therapy, nature-based therapy, outdoor and open-air therapies, forest bathing, eco-therapy, bush crafting and so on.

Context
- Research articles of quantitative and qualitative methodology, including: randomised controlled trials, controlled (non-randomised) clinical trials, controlled pretest-posttest design, prospective and retrospective comparative clinical studies, non-controlled prospective and retrospective observational studies, cohort studies with before-after design, case series, case reports, qualitative studies, systematic reviews, meta-analyses, meta-syntheses, narrative reviews, mixed-methods reviews, qualitative reviews, rapid reviews and studies published as master or bachelor theses.
- Research articles are limited to those published in the English, Swedish, Norwegian, German and the Dutch languages.

wilderness-related therapies; and the *context* is research articles of both quantitative and qualitative methodology (see box 1).

To this end, the following research questions were raised:

1. What concepts of wilderness therapy (ie, theoretical frameworks, foundations) are presented for childhood cancer survivors?
2. Which elements (content) are incorporated into wilderness therapy for childhood cancer survivors (ie, experiential learning methods, physical movement, challenge and risk-based activity, the generating and use of metaphors, involvement with natural environments, balance of structured and unstructured time in the programme, balance of social and individual time in the programme, and different type of habitats and habitat-specific activities) and which elements have not been incorporated for childhood cancer, but may be promising?
3. Which professionals (ie, profession, qualifications) facilitate wilderness therapy for childhood cancer survivors, and what relationship have wilderness therapy programmes had with treatment institutions?
4. What benefits and risks (outcomes) are reported for wilderness therapy in childhood cancer survivors?
5. To what extent are elements of the wilderness encounter incorporated into the daily life of childhood cancer survivors, how is this incorporation influenced by their domestic situation, and how does that benefit their health in the longer-term?
6. At what stage of treatment or survivorship are wilderness therapies offered to childhood cancer survivors?
7. What is the age range of childhood cancer survivors engaging in wilderness therapy?
8. To what extent does the socio-economic situation of childhood cancer survivors affect their participation in wilderness therapy programmes, and their continued ability to engage with nature/wilderness after the programme?
9. To what extent do disabilities (including physical, sensory or intellectual impairments) of childhood cancer survivors affect their participation in wilderness therapy programmes, and their continued ability to engage with nature/wilderness after the programme?
10. What is the methodological quality of the included studies on wilderness therapy for childhood cancer survivors?
11. What are the key gaps in literature around wilderness therapy for childhood cancer survivors?
12. Are there any ethical issues or challenges identified that relate to participation of childhood cancer survivors in wilderness therapy?

Design

When reviewing and synthesising literature, there are many different potential approaches available.[40] Given the aim and research questions of our study, a scoping review was deemed the most suitable type of review method. The purpose of a scoping review is to scope a body of literature in order to clarify key concepts and definitions, identify key characteristics related to that concept, examine how research is conducted on that topic, identify knowledge gaps and identify the types of available evidence.[41] This protocol was written in accordance with the Joanna Briggs Institute Reviewers' Manual for scoping reviews[39] and the guidance for conducting systematic scoping reviews as published by Peters *et al*.[42] Results of the scoping review will be reported according to the Preferred Reporting Items for Systematic review and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR).[43] A schematic representation of the essential steps in the planned scoping review is given in figure 1.

Eligibility criteria

According to the PCC mnemonic, the following eligibility criteria for inclusion of studies were defined:

1. Describing childhood cancer survivors, meaning participants of any sex diagnosed with cancer before the age of 21. A person is defined as a cancer survivor from the moment of cancer diagnosis throughout life.[44]
2. Describing wilderness-related therapies such as adventure therapy, recreation programmes, nature-based programmes, outdoor programmes, open-air programmes, forest bathing and bush-craft, in which the role of nature has both a contextual and therapeutic premise. The therapy is directed primarily at childhood cancer survivors. No restrictions with respect to the length or extent of wilderness therapy programmes or follow-up of outcomes will be applied. Studies focusing on related topics that do not primarily evaluate wilderness and/or nature experiences, such as evaluation of hospitals gardens, physical exercise programmes and animal-assisted therapy, or which do not explicitly

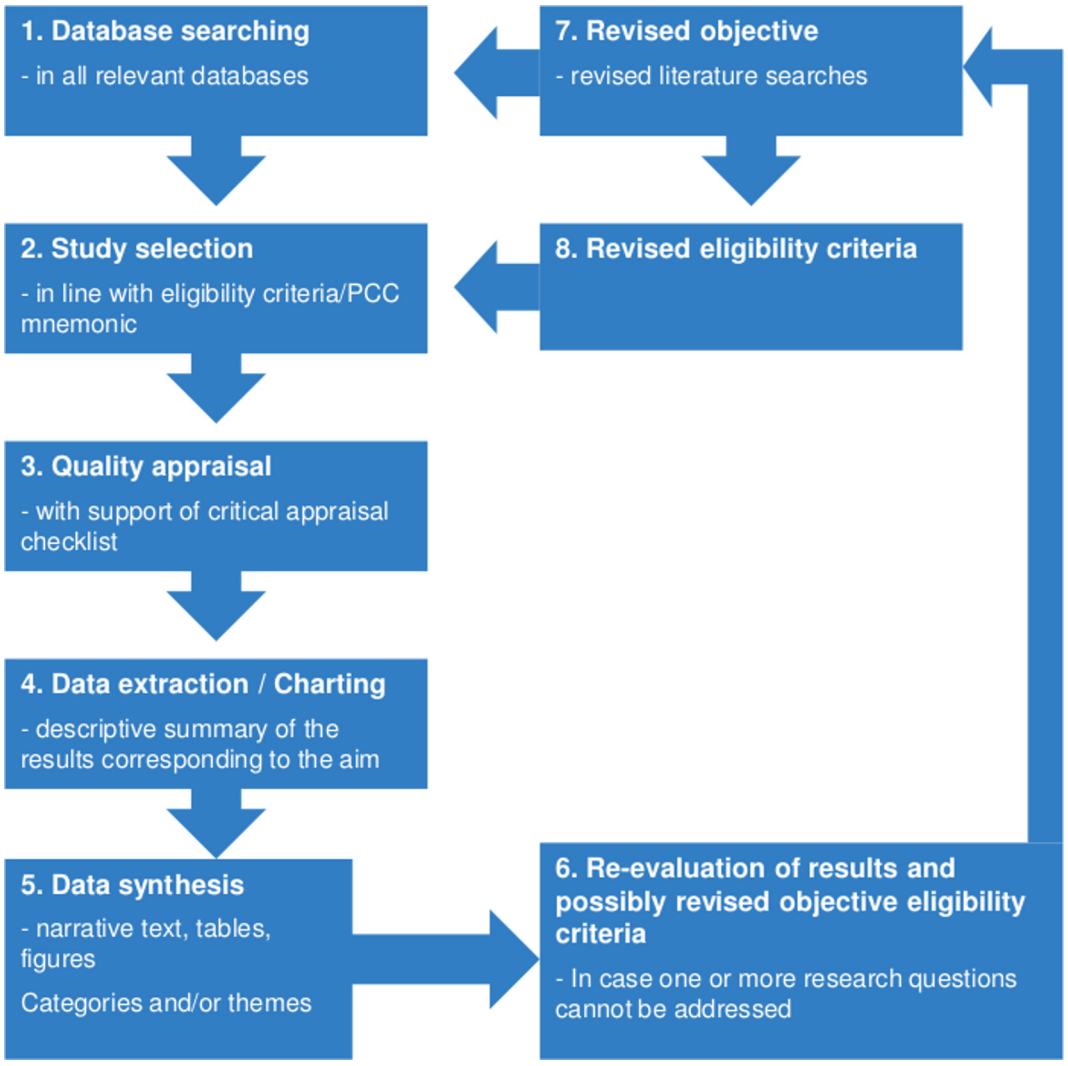

Figure 1 Schematic representation of the scoping review.

offer a programmes (individuals spending time hiking or star gazing on their own) will be excluded.

3. Research articles of quantitative and qualitative methodology, including randomised controlled trials, controlled (non-randomised) clinical trials, controlled before-after studies, prospective and retrospective comparative clinical studies, non-controlled prospective and retrospective observational studies, cohort studies with before-after design, case series, case reports, qualitative studies, systematic reviews, meta-analyses, meta-syntheses, narrative reviews, mixed-methods reviews, qualitative reviews, rapid reviews and studies published as master or bachelor theses. No restrictions by type of setting will be applied. Articles reported in the English, Swedish, Norwegian, German and Dutch languages. A list of possibly relevant articles in other languages will be provided as an appendix, provided that the abstract of these articles is available in any of the above-mentioned languages.

Research articles to be included should minimally describe: (1) A wilderness therapy programme targeted towards childhood cancer survivors, (2) Description of the content of the wilderness therapy programme and (3) At least one reported (health-related) quantitative or qualitative outcome.

Information sources and search strategy

Literature search strategies will be developed using MeSH and text words related to wilderness therapy and childhood cancer survivors. Searches will be performed in the following databases: AMED, CINAHL, Cochrane Library, EMBASE, ERIC, Google Scholar, Medline (Ovid), Psycinfo, Scopus, Sociological Abstracts, SPORTDiscus, Svemed+ and Web of Science. An MeSH search strategy was developed by two authors (WS and MCJ), one being an information specialist with expertise in systematic review searching in the field of cancer. WS will adapt the search strategy using keywords for each specific electronic database according to their specific subject headings or structure. No study design, date or language limits will be imposed on the search, although only studies in languages mentioned in the eligibility criteria will be included. The search strategies in the databases described above will be performed by one author (WS),

Box 2 MeSH terms/Text words (Title/Abstract)

Cancer
- exp Neoplasms/
- (neoplas* OR cancer* OR tumor OR tumors OR tumour* OR carcinoma* OR leukem* OR leukaem* OR malignan* OR oncolog*).ti,ab.

Wilderness therapy
- Exp Wilderness or exp Forests or exp Recreation Therapy
- (wilderness OR adventure* OR nature* OR natural OR open air* OR outdoor* OR recreational* OR forest* OR urban).ti,ab. ADJ3 (program* OR therap* OR treatment* OR rehab*).ti,ab. OR (forest bath* OR shinrin-yoku).ti,ab.

Adolescents
- exp Young Adult/ or exp CHILD/ or exp ADOLESCENT/ or exp Minors/ or PUBERTY/ or exp PEDIATRICS/
- (Child* OR Schoolchild* OR School age* OR Adoles* OR Teen* OR Boy* OR Girl* OR Minor* OR Pubert* OR Pubescen* OR Prepubescen* OR Pediatric* OR Paediatric* OR Peadiatric* OR young adult*).ti,ab.

with the exception of Svemed+ which will be performed by another author (MJ). The Medical Subject Headings and title/abstract terms for the search strategy are shown in box 2. Title/abstract terms wilderness therapy will be searched with adjacency (ADJ) operator to avoid too many irrelevant hits. The grey literature will be searched as well, as this can lead to significant contributions to a systematic review.[45] Grey literature searches will be performed by a researcher (MM, see the Acknowledgements section) in databases as recommended by the Cochrane Collaboration (PsychExtra, NTIS)[46] and Paez (Open-Grey, WONDER, CPI, PQDT Open).[45] The open-source library RT Wise Owls and the medically focused deep web search engine Mednar will be used to search conference abstracts, research publications and doctoral theses. A scoping review allows to refine or broaden the search strategy if needed.[47]

Study selection

Literature search results will be uploaded in reference management programme Endnote to facilitate the study selection process. The authors will develop and test screening questions for assessments based on the inclusion and exclusion criteria. Citation abstracts and full text articles will be uploaded into the data management programme. Prior to the formal screening process, the screening questions will be piloted and refined. A single data management file of all references identified through the search process will be produced. Duplicates will be removed from this file. These references will undergo a two-stage process of screening using the inclusion and exclusion criteria by two authors (MCJ and MJ) independently. They will screen the titles and abstracts of the searched studies, perform study selection and record their decisions on a standardised eligibility form. Disagreement between the two reviewers will be discussed with a third author (EAL) and final decisions will be made. Additional information from study authors will be sought where necessary to resolve questions about eligibility. Reasons for excluding trials will be documented. Neither of the review authors will be blind to the journal titles or to the study authors or institutions. The presentation of Results section will identify how many studies were identified and selected. A narrative description will be given of the search decision process accompanied by a flowchart of study selection and identification according to the (PRISMA-P) guidelines.[48] In this flowchart, details of the review decision process will be depicted with respect to identification, screening, eligibility and included studies. Removal of duplicate citations, reasons for exclusion and additions from a possible third search will be cited.

Data collection and charting

Two authors (MCJ and MJ) will read the articles independently, supported by two junior researchers (MM and JO) in order to extract data that will be entered in duplicate in a piloted charting form. Any disagreement between the authors will be resolved by discussion with a third author (EAL). To ensure consistency across reviewers, a pilot test will be conducted before starting the review. Data extracted will include general study information, methodology, intervention details and all reported patient-reported outcomes. The data charting process aims to generate a descriptive summary of the results corresponding to the aim and research questions of this scoping review. A concept charting form has been developed to aid the collection and sorting of key pieces of information from the selected articles (see box 3). This charting form will be piloted among the authors and possibly adapted before final data extraction is started.

Outcomes

Outcomes defined are theoretical frameworks or foundations underlying wilderness therapy in childhood cancer, the components of wilderness therapy programmes, incorporation of elements of the wilderness encounter into day-to-day life of cancer survivors, access to wilderness therapy programmes in terms of costs and disabilities, qualifications and profession of facilitators, stage of treatment or survivorship at which wilderness therapies are offered, ages of survivors to whom wilderness therapy is targeted, methodological quality of included studies, any reported short-term and long-term health-related outcome for survivors, and side-effects and other risk factors.

Quality appraisal

Although scoping reviews usually do not perform a formal quality assessment of included studies,[42] it was decided to do so in the present study in order to identify possible evidence gaps in literature. Two authors (MJ and EAL) will independently rate the methodological quality of included papers using a critical appraisal checklist. The checklist will contain subsections for different types of research articles. Qualitative studies will be assessed using the Critical Appraisal Skills Program,[49] and all

Box 3 Draft data charting form

1.Standard information
- Bibliographic information, study ID, article title, extracted by, checked by, type of study (review, RCT, qualitative study, observational study and so on), country, language, funding sources, declaration of interest.

2.Researcher details
- Authors and affiliations.

3.Aims and methods
- Study aims, objectives, methodology, methods.

4.Population
- Patients, number of patients, age, age at diagnosis, gender, type of cancer, time after cancer treatment, medications/other treatment, domestic situation, socio-economic situation, comorbidities and disabilities.

5.Type of intervention/programme
- Name, duration, setting, facilitators/trainers, costs of the programme.

6.Theoretical framework underlying the programme.
- Any described underlying framework such as theory/family systems, eclectic framework, attachment theory, family systems and so on.

7.Components of the programme
- Any component of the programme such as experiential learning methods, physical movement, challenge and risk-based activity, the generating and use of metaphors, involvement with natural environments, structured and unstructured time in the programme, social and individual time in the programme, and different type of habitats and habitat-specific activities.

8.Outcomes assessed
- Any reported short-term and long-term health-related outcome for participants such as physical, mental and social functioning, quality of life, healthcare use, re-consultations, side-effects and other risk factors, effects on and support from their family/relatives, incorporation of elements of the wilderness encounter in their day-to-day life.

9.Emergent themes
- Any theme that may be described or arise from reading the (qualitative) studies such as (for example) 'gaining control', 'feeling alive' and so on.

other studies using the critical appraisal tools from the Joanna Briggs Institute.[50] A table will be generated with the scoring for each quality item among all studies with this design. Discrepancies between the author's quality assessments will be discussed and resolved with a third author (MCJ).

Collating and summarising the results

A summary table with detailed information of every included study/source will be provided. Four authors (MCJ, EAL, HRO and MJ) will perform data synthesis. Data will be predominantly described in a narrative style, and reported in extraction tables, diagrams, and where appropriate in themes and text. Data on theoretical frameworks/foundations, the components of wilderness therapy programmes, qualifications and profession of facilitators, stage of treatment or survivorship when wilderness therapies are offered, age of survivors targeted for wilderness therapy, methodological quality of included studies, and health-related outcomes will be described and tabulated. In case one or more research questions cannot be addressed, the eligibility criteria and or search terms for inclusion of studies in this scoping review will be broadened to include young adult cancer survivors (ages 15–39 years) and/or other serious or life-threatening paediatric diseases such as immune deficiencies, blood disorders, autoimmune diseases and so on.

Ethics and dissemination

This study describes a protocol for a scoping review that will undertake secondary analysis of data already published in literature. Therefore, the present study is exempt from medical ethical review. The results of the study will be published in an international peer-reviewed journal according to the PRISMA-ScR guidelines.[43] In addition, results will be disseminated through networks of scientists, healthcare professionals and wilderness therapy practitioners, as well as at conferences.

Patient and public involvement

Since this study describes the protocol for performance of a scoping review, it was not appropriate to involve patients at this stage in this work. The research questions of this systematic review were discussed with wilderness therapy practitioners with more than 10 years' working experience with children.

DISCUSSION

The present scoping review aims to systematically map and categorise the concept, components and outcomes of wilderness therapy programmes specifically targeted at childhood cancer survivors. A priori establishment of the review protocol and subsequent publication will guide the authors in the review process, enhance methodological quality and increase transparency in how study results are obtained. The results of this scoping review are of interest to a broad audience. This includes childhood cancer survivors, their families, practitioners, clinicians and researchers with an interest in gaining better understanding of the role of wilderness therapy for childhood cancer survivors, including benefits and risks, as well as those with an interest in understanding how to build the evidence base for wilderness therapy in the future. The international study research team includes experts from the field of epidemiology, paediatric psycho-oncology, academia, clinical medicine, wilderness therapy and scientific information services. Preliminary searches for the scoping review started in April 2019. Study completion is planned in March 2020. For optimal interpretation and dissemination of the results as obtained in the scoping review other medical and experiential experts in the field of paediatric oncology and wilderness therapy will be consulted.

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Contributors All authors have contributed to the development of the review design and research questions. MCJ conceived the idea for the scoping review, contributed to the review design and development of research questions, and wrote the first draft of the protocol and manuscript. EAL contributed to the review design and development of research questions and provided detailed comments on drafts of the protocol and manuscript. HRO contributed to the review design and development of research questions and provided detailed comments on drafts of the protocol and manuscript. WS contributed to the review design and development of research questions, lead the preparation of search terms and commented on the protocol and manuscript. MJ jointly conceived the idea for the scoping review, contributed to the review design and development of research questions, jointly prepared the search terms, and commented on and revised the manuscript.

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