The use of music for children and adolescents living with rare diseases in the healthcare setting: a scoping review study protocol [version 2; peer review: 1 approved, 2 approved with reservations]

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Abstract

Background: Interest in the application of music in the health, social care and community contexts is growing worldwide. There is an emerging body of literature about the positive effects of music on the well-being and social relationships of children and adult populations. Music has also been found to promote social interaction, communication skills, and social-emotional behaviours of children with medically complex care needs. Despite significant advancements in the area, to the authors’ knowledge, this is the first scoping review to investigate the evidence for using music therapy and music-based interventions for children living with rare diseases in the healthcare setting. Therefore, the purpose of this study is to conduct a scoping review of the literature to map out the existing studies about the use of music therapy and music-based interventions with children who have rare diseases in the healthcare setting. This review will also identify gaps in current knowledge and use of these interventions.

Method: This study follows the Joanna Briggs Institute’s methodology for scoping reviews, utilising Arksey and O’Malley’s six-stage scoping review framework: 1) identifying the research question; 2) identifying relevant studies; 3) study selection; 4) charting the data; 5) collating, summarising and reporting results; and 6) consulting with relevant stakeholders step. A comprehensive search will be conducted in CINAHL Complete; MEDLINE Complete; Psychology and Behavioral Sciences Collection; and PubMed Central databases. A search strategy with selected inclusion and exclusion criteria will be used to reveal a wide range of evidence. This study will
include quantitative, qualitative and mixed research methods studies published in English from 2010 to 2020.

**Keywords**
Music, music-based intervention, music therapy, music for health, rare disease, children and adolescents, health and well-being, healthcare

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Introduction

Rare diseases affect a small number of people compared to the general population. In Europe, a rare disease is defined as a disease or disorder affecting fewer than five in 10,000 of the European population (European Commission, n.d.). Between 27 and 36 million people reside in Europe with one of the 8000 distinct diseases, which means that rare diseases create a significant public health and social care issue in Europe. Although rare diseases are individually rare, collectively, they are common in society. 50–75% of all rare diseases affect children, and the challenges faced by children and their families are often similar regardless of the specific diagnosis (Eurordis, 2009).

Children with rare diseases have multiple ongoing complex health and developmental issues due to the complex, chronic nature of rare diseases, which often cause intellectual and physical disability that place substantial demands requiring multidisciplinary input and regular hospitalisations (Anderson et al., 2013; Elliott & Zurynski, 2015; HSE, 2019; Kuo et al., 2011; vand der Lee et al., 2007). Rare disorders significantly impact children’s overall health and well-being, and development, not only physically but also psychosocially (Eurordis, 2009; HSE, 2019).

Evidence suggests that arts-based activities are effective in reducing adverse physiological and psychological outcomes. Recent findings from a review published by the World Health Organisation (Fancourt & Finn, 2019) demonstrated that the arts for health activities were used in disease prevention and health promotion (for instance, support child development, encourage health-promoting behaviours, help to prevent ill health and support caregiving) and management and treatment (e.g. support care for people with chronic conditions and many more). The “Arts on Prescription” has been used for around two decades in the United Kingdom as part of broader social prescribing schemes showing the benefits for mental health, chronic pain, management of complex and long-term conditions, social support and well-being (Drinkwater, Wildman, Moffatt, 2019 cited in Fancourt & Finn, 2019). Engagement with creative arts-based activities has been used as a vehicle to improve capacity for self-reflection and understanding of oneself and others (Stuckey & Nobel, 2010).

There is substantial evidence demonstrating the positive impact of music on an individual’s health and well-being (Hagemann et al., 2019; Impellizzeri et al., 2020; Irons et al., 2020; Ørjaseter et al., 2018; Picard et al., 2014; Tamplin et al., 2014).

There is growing interest in music therapy and music-based interventions (MT&MBI) in healthcare settings, both in adult and child populations. The use of MT&MBI has been associated with substantial psychosocial benefits. For example, exposure to music has been found to reduce stress and anxiety in pregnancy and build mother-infant relationships (Corbijn van Willenswaard et al., 2017; Wulff et al., 2015), and it has helped manage anxiety and stress (Franzoi et al., 2016; Kesselman et al., 2016) and pain associated with diagnostic procedures and treatments (Calceterra et al., 2014; Sabzevari et al., 2017; van der Heijden et al., 2015).

In children and young people, especially those living with disabilities such as autism or those who experience developmental delays, music has been shown to improve speech and communication, social interaction and emotional skills (Groß et al., 2010; Janzen & Thaut, 2018), motor function (Bharathi et al., 2019) and social outcomes (Chasenshtabar et al., 2015; LaGasse, 2017; Spiro & Himberg, 2016; Wan et al., 2010b). Other research has found benefits of music in terms of supporting well-being and coping abilities for children and young people with mental health conditions (Greboz-Haring & Thun-Hohenstein, 2018; Sollia, 2015), epilepsy (Lin et al., 2014), cancer (Ahmadi, 2013), neurological disorders (Wan et al., 2010a) and anxiety (Bibb et al., 2015), as well as building a supportive environment in the palliative care context (Archie et al., 2013; Porter et al., 2017).

Although considerable evidence indicates that music contributes to the child’s physical and mental well-being, the evidence on the use of MT&MBI for children living with rare diseases is minimal. Similarly, very little is known about how MT&MBI are used to explore the children’s and young people experiences of living with an illness.

For this reason, our cross-disciplinary team of health researchers (including arts for health researcher) decided to conduct this scoping literature review study focused primarily on the use and the impact of MT&MBI on the health and well-being of children living with a rare disease in the healthcare settings. To develop music-based interventions for this specific target group, we need to investigate the existing findings, underline the research gaps, and craft the recommendations for the researchers and policymakers to inform future research and practice.

The aim of the scoping review

The purpose of this paper is to conduct a scoping review of the literature to map out the existing studies about the use of MT&MBI with children who have rare diseases in the healthcare setting. The study also aims to explore and summarize the diversity of these studies, answering the following questions:

- What were the main characteristics of MT&MBI?
- In what healthcare settings have these studies been carried out?
• Who were the MT&MBI targeted at?
• What types of research designs methods were used?
• What theoretical frameworks were used to guide intervention development and evaluation?
• What was the nature and type of interventions?
• Which outcomes have been described in the literature?
• What were the roles and significant impacts of MT&MBI on participants’ health and well-being?

Methods
The knowledge regarding the general benefits of music on the individuals and the community's health and well-being are well-established, yet data about its impacts on children living with rare diseases remain limited. There is very little evidence on the use of MT&MBI to support health and well-being and explore lived experiences of children living with rare diseases. Thus, we have decided to conduct a scoping review, which is the optimal methodology to reflect on our study aim. It will follow Arksey & O’Malley’s (2005) original five-stage scoping review framework (identify the research question; identify relevant studies; select the studies; chart the data; collate, summarise and report the results) and more recent refinements to the method proposed by Levac et al. (2010) and the Joanna Briggs Institute (JBI) methodology for scoping reviews (Peters et al., 2020). This study will involve the following six stages in undertaking a scoping review: 1) identifying the research question; 2) identifying relevant studies; 3) selecting studies; 4) charting the data; 5) collating, summarising and reporting the results and 6) consulting with relevant stakeholders (Levac et al., 2010). This scoping review will emphasise the current evidence base. It will significantly contribute to the rare diseases research and policy focused on arts and music as a research method, effective arts-based participatory tool for research involving children, and tool to support individual-and community-level well-being.

This study protocol will outline how we will address each of the six stages.

I. Identifying the research question
In this study, we carefully planned the research question and the focus of the scoping review. To identify the research question, we searched and designed our key search terms to capture the most appropriate body of literature.

In identifying the study question, the study team identified key concepts that encompass the cross-disciplinary study’s scope. Five team members were involved in this process (including an arts for health researcher, music therapist and experienced researcher investigating the quality of life of children living with a rare disease and the care relationships between the children and their carers). The key concepts were as follows: music therapy and music interventions, health and well-being, children and adolescents living with rare diseases/conditions, healthcare setting. The study’s key focus will be MT&MBI as a research method and/or tool to support children’s health and well-being within a healthcare setting and explore their lived experiences. We will analyse the existing literature in the field to answer if and how diverse forms of MT&MBI are being used in healthcare settings for children living with rare diseases.

The study question of our research is: “What are the roles and benefits of MT&MBI as a tool for researching children living with a rare disease in healthcare settings?”

II: Identifying relevant studies
In the beginning, three researchers (including a music therapist) identified the databases for the literature search process. A comprehensive search will be conducted in the following databases: CINAHL Complete; MEDLINE Complete; Psychology and Behavioral Sciences Collection; and PubMed/PMC. These sources include esteemed journals in the area of health and social care. Additionally, if the number of articles suitable for the inclusion in the study is very small, searches will be conducted in the APA PsychINFO; ERIC; Lenus Irish Health Research Repository; and Google Advanced Search/Google Scholar databases too. The inclusion criteria will be based on the population–concept–context (PCC) framework recommended by the JBI (Peters et al., 2020). Discussion among all research team members regarding exclusion and inclusion criteria at the start of the scoping review process occurred, and two researchers made the final decision. Table 1 offers an overview of the selection of the articles (eligibility) criteria for this study.

Table 1. Overview of articles selection criteria.

| Aspects of articles selection criteria | Articles inclusion criteria | Articles exclusion criteria |
|---------------------------------------|-----------------------------|-----------------------------|
| Population                            | Human; Children living with rare diseases; Studies working with participants aged between four and 16 years. | Any other population; Any other group of participants; Any other age group. |
| Target group characteristics          |                             |                             |
|                                       |                             |                             |
III. Search strategy and study selection

Following Arksey & O’Malley’s (2005) framework, we will outline the vocabulary we will apply within our search strategy to find the most valuable literature relevant to our study question. In designing our search strategy, our team (with interdisciplinary and cross-disciplinary experience) will discuss the areas of teamwork, agree on discussions (in the main steps of the article writing process) and joint decisions, as noted by Levac et al. (2010).

To select the final keywords, we will carry out an initial literature search in a relevant database to see what definitions are dominant (searching article titles, abstracts, keywords relating to our study aims). At the end, a comprehensive set of keywords will be identified and used in our study. During the search phase, if any additional significant keywords are identified, we will include these in our searches across all databases. At the end, our search strategy will be carefully developed by our research team and if needed, in consultation with the librarian. Experts will approve carefully selected keywords. At least three researchers (including arts for health researcher, music and art therapists) will be involved in the search strategy design and listing the keywords relevant to music-focused research. The literature search strategy will be carried out using several search terms; keywords are overviewed in Table 2.

Table 2. The literature search strategy: keywords.

| Aspects of articles selection criteria | Articles inclusion criteria | Articles exclusion criteria |
|---------------------------------------|----------------------------|-----------------------------|
| **Type of activity**                  | Discussion or application of music therapy and music-based interventions, technique/session | Other art forms |
| **Context**                           | Healthcare setting (at any location both locally and internationally) | Other settings |
| **Study location**                    | Studies from peer-reviewed journals | 1) Non-peer reviewed documents and grey literature (such as book chapters, book reviews, policy reports, bibliographies, technical reports, conference proceedings and reports, accessible dissertations and theses, research protocols and reports, commentaries, editorials and letters); 2) Duplications. |
| **Accessibility**                     | Open access and full-text publications | Restricted access publications (paid) |
| **Year of publication**               | Articles published between January 2010 and October 2020 | Any publication outside these years |
| **Language**                          | Publications are written in English | Any other language |
| **Research methods**                  | Papers using qualitative, quantitative, mixed research methods and case study examples; Articles using the music therapy, music-based interventions as a research method, research tool, or initiative. | Articles with very poor quality of the study design and findings presentation; Articles that focus on other arts forms. |

| Type of arts activity / art form       | Study population                           | Health condition                                      | Study setting                                                                 |
|---------------------------------------|--------------------------------------------|-------------------------------------------------------|-------------------------------------------------------------------------------|
| music OR music therapy                | child OR children OR youth OR younger OR   | chronic disease OR chronic condition OR chronic illness| healthcare OR health care OR hospital OR hospice OR clinical OR outpatient     |
| OR music activity OR music session    |   OR kid OR teen OR teenager OR adolescent OR preteen OR junior OR immature OR young | OR rare disease OR rare illness OR rare condition OR complex needs | OR inpatient OR outpatient care OR inpatient care OR rehabilitation OR recovery OR healing OR treatment |
| OR music for health OR music and health|                                            |                                                       |                                                                                |
At least three independent reviewers will screen the listed studies by title and abstract. They will subsequently conduct a full-text review based on the pre-specified inclusion/exclusion criteria relevant to our research question. The reviewers will meet at different stages of the review process to discuss/report any challenges, clarify uncertainties, make necessary refinements to the search strategy following the recommendations provided by Levac et al. (2010), and the studies selection step. All disagreements will be solved, finding a shared agreement. Should any major disagreement arise regarding the inclusion or exclusion of studies that cannot be resolved by consensus, an additional reviewer will be invited to make the final decision.

### IV. Charting the data: initial characteristics

A chart will be used for data extraction influenced by the guidelines from the JBI Reviewer’s Manual (Peters et al., 2020). This consists of particular study aspects and related questions to inform the charting process. It will support the next stage of the review process: collating, summarising, and reporting the results, identifying themes. The process of charting data will be open to integrate new characteristics of the selected studies if these will enhance the data analysis and help to reflect on our study questions. Table 3 shows a primary table of charting characteristics and associated aspects of data extraction.

| Studies characteristics | Focus on the following aspects |
|-------------------------|--------------------------------|
| Authors, Year           | Who are the authors of the study?  |
|                         | What year was the study published? |
| Journal                 | What is the title / name of the journal? |
| Country                 | Where was the study published?     |
|                         | Where was the study conducted?     |
| Target group            | Who were the participants / who was the target audience of the study: identifying group/-s and how many persons participated in the study? |
|                         | What were the characteristics of the participants (socio-demographic, health related, etc.)? |
| Aim                     | What was / were the aim /-s of the study? |
| Setting                 | Where was the study conducted (identifying the setting / context, for e.g. identifying the hospital department; was it a clinical, outpatient, rehabilitation, other setting)? |
| Activity                | What were the forms of music applied in this study?  |
|                         | What was the duration of the programme (how many sessions, how often)? |
|                         | Who was the music activity lead (art therapist, musician, artist, clinician, researcher, other professional)? |
|                         | What was the content of the music activity? |
|                         | What form /-s of music was / were used in the study? |
|                         | Was the music combined with other art forms? |
|                         | Was the music element used at one or several parts of the study? |
|                         | Did the aspects of music activity change in the study? |
| Participant recruitment | How were the participants invited to take part in the study? Was it voluntary participation or not? |
|                         | Were there any collaborations with NGOs, patient advocacy institutions, universities, other sectors? |
| Research methods        | What types of research methods were applied in the study: qualitative, quantitative, mixed, arts-based research methods? |
|                         | Was a music professional (or other specialists) part of the research team / involved in developing the study / music aspect of the study and how? |
| Ethical approval        | Was there any evidence of ethical approval? |
|                         | Were there any specific ethical considerations / issues? |
| Data analysis           | How was the data analysed: what methods were used? |
|                         | What were the parameters for data analysis? |
| Studies characteristics | Focus on the following aspects |
|------------------------|--------------------------------|
| **Main results**       | What were the main results of the study? |
|                        | Was there any focus / what was the impact of music on participants' health and well-being? |
|                        | What was the general role of the music in disease prevention, health promotion and the process of patient hospitalisation and recovery? |
|                        | What was the impact of music on the participant's lived experiences with a rare disease / disease perception? |
|                        | How have the MT&MBI been used: as a research method or as a tool? |
| **Advantages**         | What were the main advantages of the study? |
| **Limitations**        | What were the main limitations of the study? |
| **Recommendations**    | What were the recommendations and practical implications of the study? |

### V. Collating, summarising and reporting the results

The review will be conducted and reported following the guidelines suggested by JBI Reviewer’s Manual for Scoping Reviews (Peters et al., 2020) and PRISMA Extension for Scoping Reviews (Tricco et al., 2018). We will identify the relevant information according to our study aim and question, extract and chart the data, make a narrative, and summarise the findings. The PRISMA flow chart offered by Moher et al. (2009) will be used to illustrate the search strategy.

We will thematically analyse the studies’ findings, using qualitative descriptive methods to overview the literature as Levac et al. (2010) advised. Findings will be structured into thematic categories (using thematic analysis approach suggested by Braun & Clarke, 2006), and the key findings will be presented in sections such as the introduction; aim of the study and methodology; and results of the overviewed papers, including the following information:

- general characteristics of the studies;
- aim / -s and goal / -s;
- application of the MT&MBI;
- applied methods and data analysis;
- main results – the impact of MT&MBI on the participants’ health and well-being, as well as on their lived experiences with a rare disease;
- gaps in the literature and recommendations for future research and implications for the practice.

We will provide the readers with the conclusions and identify the strengths and limitations of our research. Our study will emphasise the importance of MT&MBI for children living with rare diseases in healthcare settings. It will also inform our SAMPI research project1, helping to foster new ideas in future research and practice.

### VI. Consultation

This study is a part of the SAMPI research project, which Children’s Health Ireland implements at Temple Street Children’s University Hospital (RPAC17-05). The majority of team members in this study have experience working with children living with rare diseases, complex and chronic conditions. In this study, a cross-disciplinary consultation, as a suggested element by Levac et al. (2010), plays a significant role – all parties will be invited, and those who expressed interest will be involved from the beginning – in the planning of this study, selection of keywords, identification of inclusion and exclusion criteria, selection of studies, and commenting on this study protocol. All will be invited to discuss preliminary findings, validate findings and inform ideas about future research. The SAMPI research project will include the development and implementation of the music-based intervention for children living with a rare disease in Children’s Health Ireland at Temple Street Children’s University Hospital. This will consist of consultations with stakeholders (collaboration and knowledge exchange with children living with rare diseases, families, and advocates).

Finally, the authors believe that the information gained through this scoping literature review will:

- Contribute to the knowledge in the field about the use of music therapy and music as an intervention / research method / tool in healthcare settings for children living with rare disease, chronic and complex conditions;
- Provide the evidence on the impact of MT&MBI on the health and well-being of children and adolescents living with rare diseases and their lived experiences;
- Support the planning of healthcare services, community / health and well-being support initiatives, arts for health and other cross-disciplinary projects with children living with rare, chronic and complex conditions;
- Help to better plan the research in the field, including the recommendations for the practical implications. The results will also be relevant for other fields where

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1 SAMPI research Project (2019–2021) aims to enable children and young people living with rare diseases to express their experience of living with a rare disease and help to identify the factors that enhance, inhibit and impact their lives through sand play, arts, music, photovoice and interviews.
music-based interventions / music therapy is applied, such as arts and humanities and social care.

Research ethics
Due to the nature of this study, ethical approval is not required for this scoping review. However, this study is a part of SAMPI research project where ethical approval was previously obtained (Children’s Health Ireland Research Ethics Committee, Ref no. 17.006).

Dissemination
This study includes the academic and health care sectors. The study results will be disseminated through international conferences and peer-reviewed articles.

Study status
Database searches have been completed. Titles and abstracts have been screened, and full text publications were assessed against the eligibility criteria. In the next steps, these selected papers will be thoroughly analysed and the results of the article screening-selection process will be presented in a PRISMA flow diagram inspired by Moher et al. (2009).

Discussion
What this study will add:

- This will be a first and novel scoping review to understand what types of MT&MBI in the healthcare setting for children living with a rare disease research studies exist within scientific literature;
- This study will map out and identify the methodological approaches and the impact of MT&MBI on the health and well-being of children living with a rare disease in the healthcare setting;
- The review will extend the current knowledge in the field locally and internationally;
- The study will contribute to the development of music creative modality in the overall SAMPI Research Project. This review will inform future research, practice, and policy focused on the use of MT&MBI as a research tool in children and young people’s living with a rare disease and complex illness focused research.

The preliminary strengths of the study:

- The review will include both quantitative, qualitative and mixed methods studies, as well as case examples and will explore the evidence of the use of MT&MBI in a health care setting for children living with a rare disease and its impact with a primary focus on their health and well-being and lived experiences with a disease;
- At least two researchers (including arts for health researcher and experienced music therapist) will be involved in the design of search strategy, selecting the keywords and databases;
- At least three researchers (including arts for health researcher and experienced music therapist) will be included in designing the inclusion and exclusion criteria, selecting the studies;
- The study has the inclusion and exclusion criteria to extract the relevant studies which have an impact on making the results more reliable and strengthening the theoretical base of the study;
- The development of a studies characteristics overview framework will make the presentation of the finding more systematic.

Data availability
No data are associated with this article.

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PubMed Abstract | Publisher Full Text | Free Full Text
Fran Garry
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Thank you for the opportunity to review this study protocol. The paper provides a strong case for the review in terms of the paucity of research around the use of music therapy and music-based interventions to support children with rare diseases in healthcare settings. The various steps for undertaking a scoping review are outlined. I have listed some observations and suggestions aimed at enhancing clarity for the reader below:

Methods

Identifying the research question:
- Could the authors elaborate briefly on what is meant by ‘MT & MBI as a research method’, as distinct from ‘a tool to support children’s health and wellbeing...’ and also clarify any key differences between MT & MBI?
- The study question is: ‘What are the roles and benefits of MT & MBI as a tool for researching children living with rare disease in healthcare settings?’ However, throughout the paper, the authors refer mostly to ‘impacts’, which seems more open to the inclusion of possible challenges for researchers and participants. Could the authors clarify if the focus is on positive impacts only?

Search Strategy and Study Selection:
- Could the authors clarify if preliminary searches were carried out in a specific database or databases?
- The phrase ‘..make necessary refinements to the search strategy' seems to belong in the previous paragraph where the search strategy is discussed, rather than in the paragraph referring primarily to screening and study selection. Will the reviewers trial the preliminary data extraction form (Table 3) on an initial number of studies?

Charting the data:
- Regarding the preliminary data extraction chart: ‘Table 3. Initial table of the overview of
study findings: the characteristics of the studies', could I kindly request that the authors include a citation in the revised version for Garry et al. (2020)\(^1\) in relation to the specific music focused questions presented in the chart? The full reference for this published study protocol is: Garry F, Murphy Tighe S, MacFarlane A, Phelan H. The use of music as an arts-based method in migrant health research: a scoping review protocol [version 1; peer review: 2 approved]. HRB Open Research. 2020;3(75) doi:10.12688/hrbopenres.13121.1 I wish to clarify that this refers to my own work and the work of my co-authors. The music related and project focus questions comprised our original additions to the table edited from JBI Reviewer's Manual, 11.2.7 Data extraction (Nittas et al. 2018\(^2\); Peters et al. 2017).

Collating, summarising and reporting the results:

○ The authors say that the PRISMA flow chart will be used to illustrate the search strategy. I suggest changing this to match the purpose of the flow chart as outlined correctly further on in the ‘Study Status’ section.

Preliminary strengths of the study:

○ The authors write in future tense about the selection of the inclusion/exclusion criteria but they have already stated that this process was completed in the ‘Identifying relevant studies’ section. Could this be clarified further?

Overall, this is clearly an important and well thought out study, and it is evident that the reviewers have the expertise to carry out the scoping review in accordance with the steps they have outlined.

References
1. Garry F, Murphy Tighe S, MacFarlane A, Phelan H: The use of music as an arts-based method in migrant health research: a scoping review protocol. HRB Open Res. 2020; 3: 75 PubMed Abstract | Publisher Full Text
2. Nittas V, Mütsch M, Ehrler F, Puhan M: Electronic patient-generated health data to facilitate prevention and health promotion: a scoping review protocol. BMJ Open. 2018; 8 (8). Publisher Full Text

Is the rationale for, and objectives of, the study clearly described? Yes

Is the study design appropriate for the research question? Yes

Are sufficient details of the methods provided to allow replication by others? Partly

Are the datasets clearly presented in a useable and accessible format? Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Arts-based research and migrant health and wellbeing, community music research, arts-practice research, scoping review

I confirm that I have read this submission and believe that I have an appropriate level of
expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Karen Gardner
Public Sector Research Group, University New South Wales, Canberra, ACT, Australia

The article outlines the approach to conducting a scoping review of the literature on the use of MT&MBI with children who have rare diseases in the healthcare setting, following the methods outlined by Arksey and O'Malley. Overall the approach to review is appropriate but I found a couple of areas confusing and would benefit from further details to help the reader understand how the methods will support the research to answer the research questions.

It would be useful to provide a rationale for why the search dates back only to 2010. Is there any specific reason this date was decided upon?

I found the discussion on key words and search strategy confusing. The key words for the search strategy seem to be presented in Table 2 but the rest of the paragraph discusses the processes that will be used to select these. Please clarify how these are related.

The second paragraph under the heading "V. Collating, summarising and reporting the results" seems to confuse reporting and analysing results. How will you actually analyse the data from extracted studies - you say using thematic analysis - can you elaborate?

It isn't clear to me how will your analysis will support you to answer your research questions. For example, the list of items you will report on refers to impacts of programs on participants health and well being but your research questions are much broader than this and include characteristics of MT&MBI programs; different types of interventions; outcomes etc. Will you use a framework to guide this thematic analysis so you can categorise characteristics of MT&MBI programs; different types of interventions; outcomes?

Is the rationale for, and objectives of, the study clearly described?
Yes

Is the study design appropriate for the research question?
Yes
Are sufficient details of the methods provided to allow replication by others?  
Partly

Are the datasets clearly presented in a useable and accessible format?  
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Health service research, scoping review, evaluation

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 07 Dec 2021

Simona Karpaviciute, Children’s Health Ireland at Temple Street Children’s University Hospital, Temple St, Dublin 1, Ireland

Dear Karen Gardner,

Thank you for reviewing our paper. Please find our comments to your questions:

1. The period limit was applied to reveal the present-day literature.

2. In table 2 we present the selected keywords and in the two paragraphs above the description of their selection process.

3. Yes, we will use the thematic analysis approach to analyse the data. We will apply the guidelines suggested by Braun & Clarke (2006).

4. In this protocol study, the list of research questions will help to reveal the diverse aspects of selected studies. We believe, it will be beneficial for other researchers in the field to better understand the importance and varied impacts of MT & MBI. Yes, we will use a framework to guide the thematic analysis.

Competing Interests: No competing interests were disclosed.
Julie McMullan  
Queen's University Belfast, Belfast, UK

This study is very much needed and appears to be exploring an area of support and therapy which very little is known about at this time. Much thought has been given to identify the study question with various individuals from relevant backgrounds bringing their experiences and skills to this matter. It is obvious that much thought is also being given to the methodology being used and the types of key words that will be used to conduct the searches. The level of detail is appropriate and would enable replication to be conducted with ease. The authors have endeavored to be thorough in their protocol and demonstrate a wealth of experience and knowledge to enable them to undertake such a review.

Is the rationale for, and objectives of, the study clearly described?  
Yes

Is the study design appropriate for the research question?  
Yes

Are sufficient details of the methods provided to allow replication by others?  
Yes

Are the datasets clearly presented in a useable and accessible format?  
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Public health.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.