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Preferences about place of end-of-life care and death of patients with life-threatening illnesses and their families: a protocol for an umbrella review

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ABSTRACT

Introduction For most of history, the majority of people died at home surrounded by family. However, the global scenario has progressively changed towards hospital death and more recently in some countries back again towards home, with indication that COVID-19 may have further increased the number of home deaths. It is therefore timely to establish the state-of-the-art about people's preferences for place of end-of-life care and death, to understand the full spectrum of preferences, nuances and commonalities worldwide. This protocol describes the methods for an umbrella review which aims to examine and synthesise the available evidence regarding preferences about place of end-of-life care and death of patients with life-threatening illnesses and their families.

Methods and analysis We will search for relevant systematic reviews (quantitative and/or qualitative) in six databases from inception without language restrictions: PsycINFO, MEDLINE, EMBASE, CINAHL, PROSPERO and Epistemonikos. Following the Joanna Briggs Institute (JBI) methodology for umbrella reviews, eligibility screening, data extraction and quality assessment (using the JBI Critical Appraisal Checklist) will be done by two independent reviewers. We will report the screening process using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram. Study double-counting will be reported using the Graphical Representation of Overlap for OVErviews tool. A narrative synthesis will include 'Summary of Evidence' tables to address five review questions (distribution of preferences and reasons, influencing variables, place of care vs place of death, changes over time, congruence between preferred and actual places), grading the evidence on each question using Grading of Recommendations Assessment, Development and Evaluation (GRADE) and/or GRADE-Confidence in the Evidence from Reviews of Qualitative research.

Ethics and dissemination This review does not require ethical approval. The results will be presented at conferences and published in a peer-reviewed journal.

PROSPERO registration number CRD42022339983.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This umbrella review will allow a comprehensive examination and synthesis of quantitative and qualitative evidence into one systematic review of reviews.
⇒ Presentation of results will align with guidelines in the Joanna Briggs Institute Manual for Evidence Synthesis (2021) and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 statement.
⇒ Due to limitations of the databases and sources, most included systematic reviews will inevitably focus on studies published in English.
⇒ Some overlap of primary studies could potentially impact on data synthesis and will be reported.

quality of life of a person and their family.[1] This includes the places where they live in and are cared for until the person dies. In a society that seeks to promote people's involvement and empowerment, their preferences are paramount and should be central in decision-making processes. This is particularly important when facing an illness that can no longer be cured, with limited time to live. However, preferences about dying places depend on several factors, including experiences of illness, personal determinants and environmental factors such as healthcare input and social support.[2 3]

In addition to the patient's own preferences, it is important to consider the preferences of family members. They are frequently the ones providing most of the caregiving, in particular at home, sometimes acting as care coordinators of different formal and informal services. The preferences of the patient and of family members can align but also diverge, especially with illness progression.[4]

For most of history, the majority of people died at home surrounded by family.[5] However, the global scenario has been progressively changing, particularly since the late 1980s,

INTRODUCTION

The diagnosis of a life-threatening disease brings important changes in the comfort and

when hospitals became not only an important place of healing and care provision, but also the main place of death.[6] More recently, a new trend is emerging worldwide, with several countries shifting towards dying in the community, some reporting further increases in home deaths during the COVID-19 pandemic.[7 8]

This emerging transition pattern seems to align with people's preferences for place of end-of-life care and death, as reported by population-level studies from Europe and the USA, with the latter reporting a notably high home preference.[9 10] However, when faced with a life-threatening disease and as the disease progresses, preferences may change for some.[2 3 11–14] The available longitudinal evidence, which is scarce, suggests that most would still prefer to die at home as it improves their sense of dignity, autonomy and comfort.[3 10] Avoiding the risk of COVID-19 infection in hospitals and other health institutions may be an added reason to remain at home.[12]

Considering the new patterns and challenges for end-of-life care, it is timely to establish the state-of-the-art about people's preferences for place of end-of-life care and death. A scoping search identified several systematic reviews on the matter.[10 13–16] These reviews have not yet been appraised together to understand the full spectrum of preferences, nuances and commonalities worldwide. Variations among people with different illness types or different ages (children, adolescents and adults) and differences between the preferences of patients and their family members are not fully explored. In addition, the place where one prefers to be cared may not be the same as the place where one prefers to die; these two preferences might change over time, as the illness progresses[10] and the extent to which individual preferences are ultimately met remains unclear, hence, the need for this umbrella review, which will be the first on the topic. We aim to examine and synthesise the available evidence from systematic reviews regarding preferences about place of end-of-life care and death of patients with life-threatening illnesses and their families at a global level. Such new data will contribute to identify the strengths and gaps in the scientific knowledge and to fully capture the diversity of places that are meaningful for individuals.

Review questions
The following questions will be addressed:
1. What is the full spectrum of places where people with life-threatening illnesses and their families prefer to be cared for at the end of life and/or die and what are the underlying reasons?
2. Do preferences vary according to sociodemographic and clinical variables (including illness type and age) and between patients and their family members?
3. Do preferences for place of end-of-life care and place of death differ and, if so, why?
4. Do preferences change over time and, if so, why?
5. Are preferences met and, if not, why?

METHODS AND ANALYSIS
Protocol and registration
This protocol is registered in PROSPERO (CRD42022339983). It follows the Joanna Briggs Institute (JBI) methodology for umbrella reviews[17 18] and is reported according to the Preferred Reporting Items for Systematic review and Meta-Analysis protocols 2015 statement.[19] The expected start date of this study is 01 July 2022 and the expected end date is 30 April 2023. Any amendments to the protocol will be recorded and described in the final umbrella review report.

Patient and public involvement
Patients and/or the public will not be directly involved in this study. However, representatives of patients and carers from the International Alliance of Patients' Organizations and Eurocarers have participated in the design of the protocol and will help disseminate the results.

Inclusion criteria
Types of participants
We will consider systematic reviews of studies that include patients diagnosed with any life-threatening illness, of any age, gender and race/ethnicity, and/or their family members. Types of life-threatening illnesses include, but are not limited to, any acute or chronic illness for which curative treatment may be feasible but can fail (eg, cancer, frailty, organ failure, infectious diseases). We will also consider the study participants had life-threatening diseases if the authors stated they had been followed by palliative care or hospice care services. Reviews exclusively focused on patients with diseases that are not life-threatening or healthy individuals will be excluded as they focus on a hypothetical rather than real scenario of end of life and death.

In palliative and end-of-life care, the concept of 'family' goes beyond familial-based relationships (eg, by marriage, birth, consanguinity or legal adoption) and includes other significant persons (eg, friends, neighbours or legal representatives).[4] We will adopt this approach in the definition of family members, including all of the above. Systematic reviews focused on professionals and/or formal or informal carers other than family (eg, volunteers acting on behalf of charities) will be excluded.

Phenomena of interest
We will include systematic reviews that have analysed preferences for place of end-of-life care and/or place of death of patients with life-threatening illnesses and/or their family members and that respond to at least one of our review questions. Place of end-of-life care consists of any physical location where patients with life-threatening diseases receive care towards the end of life and place of death is the physical location where they eventually die.

Context
We will consider systematic reviews that include participants from a variety of settings where people with life-threatening illnesses are cared but also those focused on a

specific care setting, including but not limited to community care, long-term care facilities, acute care settings or palliative care units/hospices.

Types of studies

We will include both qualitative and quantitative systematic reviews with or without meta-analysis, as well as comprehensive or mixed-methods systematic reviews. Authors of relevant systematic review protocols will be contacted to request final findings; if available, the review will be included. Ongoing reviews will be listed for information. In case of updated versions of published systematic reviews being available, only the latest version will be considered.

Primary research studies, narrative reviews, scoping reviews, rapid reviews and other non-systematic reviews that incorporate theoretical studies or text and opinion papers as their primary source of evidence will be excluded. Whenever necessary, authors will be contacted to clarify.

Search strategy

We will identify systematic reviews searching six electronic databases:
1. PsycINFO (from 1806 to 11 October 2022).
2. MEDLINE (from 1950 to 11 October 2022).
3. EMBASE (from 1980 to 11 October 2022).
4. CINAHL (from 1981 to 11 October 2022).
5. PROSPERO (from 2011 to 11 October 2022).
6. Epistemonikos (from 2012 to 11 October 2022).

The search strategy combines controlled vocabulary and keywords defined according to the review aim with terms to identify systematic reviews. Box 1 shows the MEDLINE search strategy (via Ovid); others are detailed in online supplemental file.

In addition, we will check the reference lists of all included reviews, and contact authors and investigators carrying out research in this area for further systematic reviews and unpublished data. A search for grey literature will aim to identify eligible systematic reviews from reports of governments and non-governmental organisations. We will search CORDIS, the primary source of results from European Commission-funded research (https://cordis.europa.eu/), and websites of the National Institute for Health and Care Research (https://www.nihr.ac.uk/) and the Agency for Healthcare Research and Quality (https://www.ahrq.gov/).

Study selection

All retrieved articles will be imported into EndNote V.X9 for removal of duplicates (automatic and manual) and screening. Two reviewers will independently screen titles/abstracts to judge eligibility. Full text of potentially relevant systematic reviews will be assessed also by two independent reviewers. We will exclude reviews without a clear description of the review question, eligibility criteria, search in at least two databases and critical appraisal using a standardised tool conducted by two reviewers

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> **Box 1 MEDLINE (via Ovid) search strategy**
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> 1. palliative or hospice* or terminal* or end of life or end stage* or supportive.mp
> 2. (advanced or life limiting or life threatening) adj (disease* or condition* or illness*).mp
> 3. exp Palliative Care/ or exp "Hospice and Palliative Care Nursing"/ or exp Palliative Medicine/
> 4. exp Terminal Care/ or exp Terminally Ill/
> 5. exp Hospices/ or exp Hospice Care/
> 6. exp Death/ or exp Attitude to Death/
> 7. 1 or 2 or 3 or 4 or 5 or 6
> 8. prefer* or choice* or wish* or decision* or decid* or choos*.mp
> 9. exp Patient Preference/
> 10. exp Choice Behavior/
> 11. exp Decision Making/
> 12. 8 or 9 or 10 or 11
> 13. place* or location* or site* or setting* or context* or where or home or hospital* or hospice*.mp
> 14. care or caring.mp
> 15. death* or dying or die*.mp
> 16. 14 or 15
> 17. 13 adj3 16
> 18. systematic review* or systematic literature review or systematic scoping review or systematic narrative review or systematic qualitative review or systematic evidence review or systematic quantitative review or systematic meta review or systematic critical review or systematic mixed methods review or systematic mapping review or meta ethnography or meta synthesis or meta aggregation or "systematic search and review" or systematic integrative review or systematic Cochrane review or meta analysis.mp
> 19. exp Systematic Review/ or exp Meta-analysis/
> 20. 18 or 19
> 21. 7 and 12 and 17 and 20

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independently, since these items are critical for inclusion. Disagreements will be resolved by a consensus and with a third reviewer when needed. Reasons for exclusion will be recorded at each screening stage. Reviewers will not take part in decisions about studies in which they were involved in.

There will be no language restrictions. The review team includes researchers fluent in English, Portuguese, Spanish and French. Other languages will be translated through https://www.deepl.com and, if necessary, resorting to colleagues fluent in the language in question or professional translators.

Assessment of methodological quality

The selected systematic reviews will be critically appraised independently by two reviewers. For this purpose, we will apply the standard JBI Critical Appraisal Checklist for Systematic Reviews and Research Syntheses (table 1)[17] and follow the JBI Reviewers' Manual 2021.[18] Disagreements will be solved by a consensus and with a third reviewer when needed. Whenever necessary, authors of the reviews will be contacted to request missing or additional information. Reviewers will not assess the methodological quality of studies in which they were involved in.

Table 1 JBI Critical Appraisal Checklist for Systematic Reviews and Research Syntheses[17]

		Yes	No	Unclear	Not applicable
1.	Is the review question clearly and explicitly stated?	☐	☐	☐	☐
2.	Were the inclusion criteria appropriate for the review question?	☐	☐	☐	☐
3.	Was the search strategy appropriate?	☐	☐	☐	☐
4.	Were the sources and resources used to search for studies adequate?	☐	☐	☐	☐
5.	Were the criteria for appraising studies appropriate?	☐	☐	☐	☐
6.	Was critical appraisal conducted by two or more reviewers independently?	☐	☐	☐	☐
7.	Were there methods to minimise errors in data extraction?	☐	☐	☐	☐
8.	Were the methods used to combine studies appropriate?	☐	☐	☐	☐
9.	Was the likelihood of publication bias assessed?	☐	☐	☐	☐
10.	Were recommendations for policy and/or practice supported by the reported data?	☐	☐	☐	☐
11.	Were the specific directives for new research appropriate?	☐	☐	☐	☐

JBI, Joanna Briggs Institute.

To obtain an overall appraisal of each review, the following quality thresholds were defined: low quality (0%–49% of criteria met), moderate quality (50%–74% of criteria met) and high quality (75% or more of criteria met). Results of the quality appraisal will be presented visually using a traffic light scheme to display the score of each item in each review (green for 'yes', red for 'no', yellow for 'unclear', with blank for 'not applicable'). The overall methodological quality will be described in the final report.

Data collection

Data from each included systematic review will be extracted by two independent reviewers using a predesigned data extraction form. The following information will be extracted: authorship and funding sources, year of publication, type of review, existence of protocol, databases included in the search, search time frames, search strategy used, additional sources and resources searched, number of studies included in the review and their design, countries where the primary studies were conducted, study participants included (number, sociodemographic and clinical characteristics including age and illness types, distinguishing patients and family members), quality assessment methods including the instruments used to appraise the primary studies, analysis methods, findings on preferences for place of end-of-life care and death (quantitative and/or qualitative), separately for each of our review questions.

Before the data collection starts, the data extraction form will be piloted independently by two reviewers to ensure the questions are interpreted in the same way. Disagreements will be solved by a consensus and with a third reviewer when needed. Whenever necessary, authors of the reviews will be contacted to request missing or additional information. Reviewers will not extract data for studies in which they were involved in.

Data summary

We will report the screening process using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram.[20] Study double-counting in different systematic reviews will be presented visually using the Graphical Representation of Overlap for OVErviews (GROOVE) tool.[21] GROOVE provides a matrix of evidence with the number of included primary studies and systematic reviews, determining the number of overlapped and non-overlapped primary studies. The tool also allows calculation of the overall corrected covered area (CCA) using a formula (with variables from the matrix) that quantifies the overlap degree. According to the authors, a CCA of 0%–5% represents a slight overlap, 6%–10% a moderate overlap, 11%–15% a high overlap and above 15% a very high overlap.

The key characteristics of all included reviews will be tabulated, accompanied by a narrative synthesis in text of the body of evidence. Findings on our review questions will then be narratively presented, identifying the reviews that address each (number and reference) and supported by tables. A main 'Summary of Evidence' table will list all preferred places, with total preference estimates per review (as presented by their authors, for example, pooled % or range between studies) and reasons underlying preferences for each place, based on qualitative evidence (review question 1). We will subanalyse preferences of child, adolescent and adult patients, if possible. Findings will be separated for patients and family members, highlighting reviews that summarise dyadic data. Factors found to influence preferences (eg, illness type) will be tabulated, with review references, association direction and estimates if possible (question 2). We will separate and compare findings on preferences for place of care and for place of death, highlighting reviews that summarise paired data for the same individuals, and narratively synthesising qualitative evidence on reasons for discrepancies

(question 3). Estimated percentages of people who change preference over time and the direction of changes will be presented, accompanied by qualitative evidence on underlying reasons (question 4). Estimates of congruence between preferred and actual places will be presented, together with evidence on underlying reasons for gaps (question 5). Any other important findings that emerge from the reviews will be narratively reported.

We will grade the evidence on each review question using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach for quantitative evidence[22] and GRADE-Confidence in the Evidence from Reviews of Qualitative research for qualitative evidence.[23] We will explore clues of selective reporting of studies (ie, publication bias, for example, examining strategies used to find unpublished studies) or of results within studies (ie, selective reporting bias, for example, comparing the protocol with the final review).

In the discussion section of our final report, quantitative findings will be compared and integrated with qualitative findings to address each review question and deepen understanding of what the different places mean for individuals.

ETHICS AND DISSEMINATION
The study is an umbrella review, which requires no ethical approval. We will present the results at conferences and publish the final report in a peer-reviewed journal.

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